

NEURO-EDUCATION AND NEURO-REHABILITATION

EDITED BY : Eduardo Martínez-Montes, Julie Chobert and Mireille Besson
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NEURO-EDUCATION AND NEURO-REHABILITATION

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In the last decade, important discoveries have been made in cognitive neuroscience regarding brain plasticity and learning such as the mirror neurons system and the anatomo-functional organization of perceptual, cognitive and motor abilities.... Time has come to consider the societal impact of these findings. The aim of this Research Topic of Frontiers in Psychology is to concentrate on two domains: neuro-education and neuro-rehabilitation. At the interface between neuroscience, psychology and education, neuro-education is a new inter-disciplinary emerging field that aims at developing new education programs based on results from cognitive neuroscience and psychology. For instance, brain-based learning methods are flourishing but few have been rigorously tested using well-controlled procedures. Authors of this Research Topic will present their latest findings in this domain using rigorously controlled experiments. Neuro-rehabilitation aims at developing new rehabilitation methods for children and adults with

learning disorders. Neuro-rehabilitation programs can be based upon a relatively low number of patients and controls or on large clinical trials to test for the efficiency of new treatments. These projects may also aim at testing the efficiency of video-games and of new methods such as Transcranial Magnetic Stimulation (TMS) for therapeutic interventions in children or adolescents with learning disabilities.

This Research Topic will bring together neuroscientists interested in brain plasticity and the effects of training, psychologists working with adults, with normally developing children and children with learning disabilities as well as education researchers directly confronted with the efficiency of education programs. The goal for each author is to describe the state of the art in his/her specific research domain and to illustrate how her/his research findings can impact education in the classroom or rehabilitation of children and adolescents with learning disorders.

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Editorial: Neuro-Education and Neuro-Rehabilitation

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Keywords: learning disorders, language, musical training, sensorimotor training, neurofeedback

The Editorial on the Research Topic

Neuro-Education and Neuro-Rehabilitation

The latest advances in cognitive psychology, neuropsychology, and cognitive neuroscience has brought from science fiction to reality the possibility of influencing our brain activity (Owen et al., 2010; Glannon, 2014; Gruzelier, 2014a). Better understanding of brain functioning and brain plasticity has allowed neuroscientists to transfer findings from fundamental research to education and to the rehabilitation of learning disabilities (Besson et al., 2011; Goswami, 2016). The emerging fields of neuro-education and neuro-rehabilitation aim at creating effective and safe programs to improve brain functioning related to specific perceptive, cognitive, emotional, and motor abilities. Some attempts to achieve these goals take advantage of the use of natural mechanisms, such as those mediating the interactions between brain and arts (Särkämö et al., 2008; Bringas et al., 2015). Others use experimental designs to make the brain aware of its own activity, creating the so-called neurofeedback loop (Gruzelier, 2014b). Succeeding in these goals would constitute an achievement of high societal impact (Davidson and McEwen, 2012; Vuilleumier et al., 2014).

This *Frontiers Research Topic* brings together 16 articles that cover a broad scope of topics in the relatively young but very dynamic fields of neuro-education and neuro-rehabilitation. Contributed by world-renowned scientists in cognitive psychology, neuropsychology, and cognitive neuroscience, often experts in different types of learning disorders, this E-book is organized around three main themes: neuro-education, neuro-rehabilitation and basic research with relevance to both fields. Each theme includes Review articles covering the state-of-the-art of knowledge in a specific sub-domain, Original Research articles reporting new discoveries and Opinion and Hypothesis and Theory articles adding exciting new ideas and approaches for neuro-educational and neuro-rehabilitation methods.

In the first part, dedicated to neuro-education, Ylinen and Kujala review the impact of auditory or phonological training on the level of performance in various tasks and on the neural basis of behavior in children with dyslexia, children with specific language impairment and children with language-learning impairment. François et al. review the efficacy of musical training for language learning. They highlight several studies showing that learning to play a musical instrument can induce substantial neuro-plastic changes in cortical and subcortical regions of motor, auditory and speech processing networks. They show evidence that musical training can be an alternative, low-cost and effective method for the treatment of language-learning impaired populations, as well as for patients with stroke or Parkinson Disease. Direct support for the use of musical training for the rehabilitation of children with dyslexia is reported by Habib et al. who tested the efficacy of a specially-designed Cognitive-Musical training (CMT) method. Intensive short-term CMT with dyslexic children yielded significant improvements in categorical and auditory perception

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of temporal components of speech, while long-term CMT provided additional improvements in auditory attention, phonological awareness, reading abilities and repetition of pseudo words. Along the same lines, Fonseca-Mora et al. also tested the efficacy of a new phonological training program (with and without music), for teaching to read in a foreign language, demonstrating its beneficial effects on early reading skills but without additional improvements linked to music training. Final but not least, Kraus et al. present the results of a longitudinal study examining the impact of a community music program on language development in children from low socio-economic backgrounds. Children more engaged in the music program developed stronger brain encoding of speech and improved reading scores, thereby suggesting that this kind of program provides children with auditory enrichments that may counteract some of the biological consequences of growing up in poverty. To conclude this section, the reader will find a novel approach to neuro-education, as proposed by Gerdes et al. They view learners in terms of their neurodevelopmental trajectories and propose a groundwork for allostatic neuro-education (GANE). Illustrative case studies of the use of GANE in children with Asperger's syndrome, attention-deficit hyperactivity disorder and reading difficulties are presented.

The second part of this E-book is dedicated to neuro-rehabilitation. Serniclaes et al. review the efficacy of remediation methods that tap into core deficits in dyslexia (phonemic, grapho-phonemic, and graphemic) and examine how some of these methods may contribute to the remediation of allophonic perception. Guilbert et al. describe different procedures for sensory training in unilateral spatial neglect (USN) and present recent scientific evidence that makes music a good candidate for USN patients' rehabilitation. Dhami et al. consider the use of dancing as an intervention tool and as a potential parallel to physical and music therapies, since dancing also engages various perceptive, cognitive, emotional and motor functions. The opinion paper from Stahl and Kotz addresses three relevant issues in current research on singing and aphasia: articulatory tempo, clinical research designs and formulaic language resources. The authors discuss how these issues may reconcile seemingly contradictory findings in the literature and provide guidelines for future research based on holistic and analytic approaches that may help improving the efficacy of music-based aphasia therapy. Finally, in a Hypothesis and Theory article, Elmer and Jäncke consider the use of a neurofeedback approach for auditory rehabilitation. They first stress the advantages of

using intracerebral functional connectivity (IFC) instead of quantitative EEG for interventional applications and then propose concrete interventional IFC applications that may improve auditory-related dysfunctions such as developmental dyslexia.

In the third part, we compiled some interesting studies which contribute both to a better comprehension of basic psychophysiological mechanisms and to the development of potential applications for neuro-education and neuro-rehabilitation. Vidal et al. review the role of sensorimotor information for motor learning. They discuss the effects of several factors known to influence information processing in sensorimotor activities based on the distinction between extrinsic (e.g., quantity and quality of information, level of instruction and motor program learning) and intrinsic factors (e.g., prior information, individual strategies and capabilities for fast error detection). In a more specific context, Berteletti and Booth investigated the extent to which somatosensory information from the fingers contributes to numerical sense in children. Their work provided first neurological evidence for a functional role of the somatosensory finger area in proficient arithmetic problem solving, thereby encouraging educational practices to integrate finger-based strategies as a tool for instilling stronger numerical sense. Still in another context, writing, Danna and Velay review studies that use natural sensory and supplementary feedback to help the writer learn how to write and to control writing. They discuss the role of each sensory modality, how information is used in handwriting control and how this control changes with practice and learning. Turning from writing to reading, Boudelaa reports, in an original research article, that the processing time course in auditory modality is different for consonants and vowels in Arabic. The implications of this work for neuro-education and neuro-rehabilitation in Arabic are discussed. Finally, Tandonnet et al. illustrate how basic approaches in cognitive science may benefit human factors engineering and potentially improve man-machine interfaces.

We hope this compilation of articles describing the latest research in the field of neuro-education and neuro-rehabilitation will be of interest to the readers and will impulse even more research in these fascinating new fields with strong societal impact.

AUTHOR CONTRIBUTIONS

All authors listed, have made substantial, direct and intellectual contribution to the work, and approved it for publication.

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Neuroscience illuminating the influence of auditory or phonological intervention on language-related deficits

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Remediation programs for language-related learning deficits are urgently needed to enable equal opportunities in education. To meet this need, different training and intervention programs have been developed. Here we review, from an educational perspective, studies that have explored the neural basis of behavioral changes induced by auditory or phonological training in dyslexia, specific language impairment (SLI), and language-learning impairment (LLI). Training has been shown to induce plastic changes in deficient neural networks. In dyslexia, these include, most consistently, increased or normalized activation of previously hypoactive inferior frontal and occipito-temporal areas. In SLI and LLI, studies have shown the strengthening of previously weak auditory brain responses as a result of training. The combination of behavioral and brain measures of remedial gains has potential to increase the understanding of the causes of language-related deficits, which may help to target remedial interventions more accurately to the core problem.

Keywords: language deficit, dyslexia, neuroscience, training, remediation, intervention

INTRODUCTION

Finding the most effective techniques to remediate language-related impairments, such as dyslexia, specific language impairment (SLI), or language-learning impairment (LLI, cf. Tallal, 2001), would be of crucial importance to educators, who try to help children struggling with these learning difficulties. This raises a question, whether understanding the neurobiological underpinnings of language impairments facilitates their efficient treatment. In this review, we discuss how neuroscience illuminates the effects of auditory or phonological intervention on dyslexia, SLI, and LLI. We focus on auditory or phonological interventions, because in many cases dyslexia, SLI, and LLI are all characterized by phonological (or auditory) deficits (Tallal, 2001; Shaywitz and Shaywitz, 2005; Pennington and Bishop, 2009; Ramus et al., 2013), despite their complex etiology. Whereas detailed brain areas influenced by reading interventions can be found in a recent meta-analysis by Barquero et al. (2014), here we address whether neuroscientific research on the remediation of language-related deficits is useful for educators and whether it has something to add over behavioral research from an educational perspective.

In the current review, the selection of publications was based on the following criteria: the research should concern dyslexia, SLI, or LLI, include testing before and after an auditory or phonological intervention or training, involve brain research measures [(functional) magnetic resonance imaging (MRI/fMRI), magnetic source imaging (MSI) or magnetoencephalography (MEG), event-related potentials (ERP), or electroencephalography (EEG)], and compare two or more groups of participants to control for the effects of repeated testing and maturation (McArthur, 2009). Searches from Web of Science and PubMed (keywords dyslexia/SLI/LLI,

intervention/remediation/training, fMRI/MEG/ERP) were used in finding literature. Additional publications were found in the reference lists of relevant studies.

IS NEUROSCIENTIFIC RESEARCH USEFUL FOR EDUCATORS?

Research on remedial interventions for learning deficits may have important applicability to education (Tallal, 2012). In this area, collaboration between education and neuroscience could result in mutual benefits (Sigman et al., 2014). However, the value of the neuroscientific approach in such research has been questioned by Bishop (2013) because of methodological and interpretive reasons. She argued that neuroscientific studies often use small subject groups, which may decrease their reliability and result in small statistical power (cf. Button et al., 2013). Furthermore, Bishop (2013) noted that some studies lack an adequate control group, which is important to control for the effects of repeated testing and maturation (see also McArthur, 2009). Indeed, future intervention studies should not only aim at having larger subject groups (Bishop, 2013) and adequate control groups (McArthur, 2009; Bishop, 2013), but also control for placebo effects (Boot et al., 2013).

Bishop (2013) also argued that the critical test of the effectiveness of interventions is the change of behavior rather than that of brain function; changes in the brain should not be considered more important than changes in behavior. However, rather than emphasizing the brain over behavior, neuroscientific intervention studies typically aim to determine the links between brain function and behavior. Importantly, understanding the link or correlation between brain activation and skills as a result of training may help to explain how and why remedial gains take place. Since the combination of neuroscientific and behavioral measures has been

shown to be a better predictor of reading skills than behavioral measures alone (Hoeft et al., 2007; Maurer et al., 2009), this combination has potential to outperform mere behavioral measures in the study of remedial gains. Cognitive neuroscience has, in our opinion, also some advantages over behavioral research that were not mentioned by Bishop (2013). Especially when working with children whose motivation and skills can affect their performance considerably, a possibility to study the effects of intervention without subject's active effort or attention is a clear advantage. This is possible, for example, by recording mismatch negativity (MMN) brain response (Näätänen et al., 2007; Kujala and Näätänen, 2010).

From educators' perspective, neuroscientific research is seldom directly applicable in the assessment of remedial interventions. Importantly, however, educators may benefit from neuroscientific research by obtaining a more detailed picture of relevant processes underlying behavior. For example, brain measures may help to disentangle whether behaviorally observed improvement is due to the normalization of the core deficit or some compensatory strategy (e.g., Eden et al., 2004; Shaywitz et al., 2004), which is not evident in behavioral data. If, hypothetically, some intervention resulted in the formation of a compensatory function to solve some task, it may improve behavior to a certain degree but might not compete in effectiveness with the optimal function for solving that task. Still, in a large subject group, this compensatory improvement in behavior may be taken to reflect a successful intervention, if statistically significant improvement is achieved. Thus, neuroscientific research can potentially give some valuable information to educators about the deficits, which may help to target the contents of interventions more accurately.

OVERVIEW OF STUDIES ON NEUROBIOLOGICAL CHANGES FOLLOWING PHONOLOGICAL OR AUDITORY INTERVENTIONS

As shown by **Tables 1** and **2**, the majority of studies on phonological or auditory interventions focused on dyslexia or related problems in reading, writing, or spelling. Furthermore, the majority of studies have focused on children. Older age groups should not be neglected in remediation and its research, however: as noted by Eden et al. (2004), most dyslexics are adults, who may suffer from the socio-economic consequences of their reading deficit. There seem to be no constraints with respect to brain plasticity that would hinder remediation in adults or older children (Simos et al., 2002; Eden et al., 2004). Nevertheless, the earlier the interventions are conducted, the more benefit to individuals is gained, because learning is cumulative. The early gains may help to prevent difficulties not only in academic but socio-emotional domain. The optimal timing of intervention is, however, determined by maturity and acquired skills. For example, if a new skill is scaffolded by previous skills, it cannot be adapted before they are mastered (cf. Jolles and Crone, 2012).

The studies listed in **Tables 1** and **2** suggest that in addition to behavior, the remedial gains of phonological or auditory interventions are consistently reflected in different aspects of brain functioning. These include increased or normalized brain activation as a result of training in previously hypoactive areas as measured with fMRI (Aylward et al., 2003; Temple et al., 2003; Eden et al., 2004; Shaywitz et al., 2004; Gaab et al., 2007; Meyler et al., 2008; Heim et al., 2014) and MSI or MEG (Simos et al.,

2002; Pihko et al., 2007) during different cognitive tasks. MRI-based proton MR spectroscopy has shown normalized metabolism in certain brain areas after interventions (Richards et al., 2000, 2002). Training-induced changes in strength and timing of neural responses to stimulation have been demonstrated with ERPs (Kujala et al., 2001; Hayes et al., 2003; Stevens et al., 2008, 2013; Jucla et al., 2010; Lovio et al., 2012; Hasko et al., 2014). Also the time-frequency analysis of EEG has revealed amplitude increases in the oscillatory brain activity after training (Heim et al., 2013). In addition to brain function, interventions have been found to change brain anatomy, such as white matter integrity (Keller and Just, 2009). **Tables 1** and **2** also show that remedial gains, if any, consistently manifest in both behavioral and brain measures: in 16 out of 17 studies of **Table 1** and in all four studies of **Table 2**, remedial gains were found in both brain activation and skills targeted by intervention (note that Jucla et al., 2010, failed to find different behavioral improvement and similar brain response patterns between their treatment group and controls). The strong coupling of training gains in behavior and brain activation suggests that most likely the observed changes in the brain drive the changes in the behavior. As neuroscientific research may reveal the neural dynamics of processes related to behavioral performance and allows localize the deficient brain functions, it may enable to specify the neural mechanisms underlying language-related impairments and to determine brain functions and areas altered by interventions, which surface in behavior as improved skills. However, it is noteworthy that **Tables 1** and **2** lists published studies, whereas studies failing to find changes in behavior or brain activation may remain unpublished. This may cause bias toward systematically finding the coupling between neural and behavioral gains.

A recent meta-analysis of neuroscientific research exploring reading networks in the brain has suggested that dyslexia is characterized by the dysfunction of left occipito-temporal cortex, left inferior frontal gyrus, and the inferior parietal lobule (Richlan, 2012; see also Richlan et al., 2011). These brain areas are involved in phonological encoding, phonological representations, and attention, respectively (Richlan, 2012). Barquero et al.'s (2014) meta-analysis of the neuroimaging of reading interventions, in turn, suggests intervention-induced functional changes in the left thalamus, left middle occipital gyri, bilateral inferior frontal gyri, right insula, and right posterior cingulate gyrus. Thus, both Richlan's (2012) and Barquero et al.'s (2014) findings point toward the central role of inferior frontal and occipito-temporal/occipital dysfunction in dyslexia. Correspondingly, the neuroscientific dyslexia studies included in **Table 1**, involving auditory or phonological intervention, have shown normalized brain activation, metabolism, or anatomy as a result of interventions in the occipito-temporal (Aylward et al., 2003; Heim et al., 2014) and inferior frontal (Richards et al., 2000, 2002; Aylward et al., 2003; Shaywitz et al., 2004; Heim et al., 2014) areas. In addition, normalized activation following interventions has been repeatedly observed in inferior parietal (Temple et al., 2003; Eden et al., 2004; Meyler et al., 2008, see also Richlan, 2012), superior parietal (Aylward et al., 2003; Eden et al., 2004; Meyler et al., 2008), and temporal (Simos et al., 2002; Aylward et al., 2003; Temple et al., 2003; Shaywitz et al., 2004) areas. Although inferior frontal and

Table 1 | Publications including neuroscientific research on phonological or auditory remediation of dyslexia (or its risk).

Reference	Age of participants (years; mean or range)	Participant <i>N</i> (treatment; control)	Impairment or problem	Content of training	Duration of training	Brain research method	Task in testing	Behavioral improvement (pre-test vs. post-test)	Normalization of brain activation
Aylward et al. (2003)	11	10; 11	Dyslexia	Linguistic awareness, alphabetic principle, fluency, reading comprehension	2 weeks (28 h)	fMRI	Phoneme mapping, morpheme mapping	Yes	Yes
Eden et al. (2004)	41–44	19; 19	Dyslexia	Sound awareness, establishment of the rules for letter-sound organization, sensory stimulation, articulatory feedback	8 weeks (112 h)	fMRI	Repeating words, sound deletion	Yes	Yes
Gaab et al. (2007)	10	22; 23	Dyslexia	FastForWord*	8 weeks (about 67 h)	fMRI	Pitch discrimination	Yes	Yes
Hasko et al. (2014)	8	28 (11 improvers, 17 non-improvers); 25	Dyslexia	Phoneme discrimination and orthographic knowledge; phonics training	6 months (30 h)	ERP	Phonological lexical decision	Yes (improvers); no (non-improvers)	Yes (improvers); no (non-improvers)
Heim et al. (2014)	8–11	35 (12 training phonology, 7 training attention; 14 training reading); 10	Dyslexia	Phonological (Würzburger Trainingsprogramm, Kieler Leseaufbau), attentional (CogniPlus, Celeco), reading (Blitzschnelle Worterkennung)	4 weeks (10 h)	fMRI	Reading	Yes	Yes
Juola et al. (2010)	9–11	24; 10	Dyslexia	Phonological training; visual and orthographic training	2 months (about 16 h)	ERP	Visual lexical decision	Yes (but also in controls)	Mixed (treatment group showed a different pattern than controls)

(Continued)

Table 1 | Continued

Reference	Age of participants (years; mean or range)	Participant <i>N</i> (treatment; control)	Impairment or problem	Content of training	Duration of training	Brain research method	Task in testing	Behavioral improvement (pre-test vs. post-test)	Normalization of brain activation
Keller and Just (2009)	8–10	35 treated poor readers; 12 non-treated poor readers; 25 non-treated good readers	Poor reading	Corrective Reading, Wilson Reading, Spell Read Phonological Auditory Training, Failure Free Reading	6 months (100 h)	DTI	–	Yes	Yes
Kujala et al. (2001)	7	24; 24	Dyslexia	Non-linguistic audiovisual matching	7 weeks (about 3 h)	ERP	Passive listening, attention directed elsewhere	Yes	Yes
Lovio et al. (2012)	6–7	10; 10	Difficulties in reading-related skills	GraphoGame: letter-sound correspondences (vs. number-knowledge game for controls)	3 weeks (3 h)	ERP	Passive listening, attention directed elsewhere	Yes	Yes
Meyler et al. (2008)	10	23; 12	Poor reading	Corrective Reading, Wilson Reading, Spell Read Phonological Auditory Training, Failure Free Reading	6 months (100 h)	fMRI	Sentence comprehension	Yes	Yes
Richards et al. (2000)	10–13	8; 7	Dyslexia	Phonological and morphological reading instruction	3 weeks (30 h)	Proton MR spectroscopy	Phonological and lexical access and a non-linguistic tone task	Yes	Yes

(Continued)

Table 1 | Continued

Reference	Age of participants (years; mean or range)	Participant <i>N</i> (treatment; control)	Impairment or problem	Content of training	Duration of training	Brain research method	Task in testing	Behavioral improvement (pre-test vs. post-test)	Normalization of brain activation
Richards et al. (2002)	9–12	10; 8	Dyslexia	Phonological vs. morphological reading instruction	3 weeks (30 h)	Proton MR spectroscopy	Phonological and lexical tasks, passive listening	Yes	Yes
Shaywitz et al. (2004)	6–9	37 (experimental intervention); 12 (community intervention); 28 (control)	Reading disability	Phonological intervention: sound-symbol associations, phoneme analysis, timed reading, oral story reading, dictation (vs. community intervention in school)	8 months (50 min/day)	fMRI	Cross-modal letter identification	Yes (experimental group); no (community intervention)	Yes (experimental group); no (community intervention)
Simos et al. (2002)	7–17	8; 8	Dyslexia	Phono-Graphix (phonological processing and decoding), Lindamood Phonemic Sequencing	2 months (80 h)	MSI	Pseudoword rhyme-matching	Yes	Yes
Stevens et al. (2013)	5	8; 6	Risk for reading disability	Early Reading Intervention (phonemic awareness, alphabetic understanding, letter writing, word reading, spelling, sentence reading)	8 weeks (20 h)	ERP	Selective auditory attention	Yes	Yes
Temple et al. (2003)	8–12	20; 12	Dyslexia	FastForWord*	8 weeks (about 47 h)	fMRI	Rhyme letters, match letters, match lines	Yes	Yes

*DTI, diffusion tensor imaging; ERP, event-related potential; fMRI, functional magnetic resonance imaging; MR, magnetic resonance; MSI, magnetic source imaging. *FastForWord includes auditory discrimination, phoneme discrimination, phoneme identification, phonic match, phonic word, understanding instructions, grammatical structures and rules.*

Table 2 | Publications including neuroscientific research on phonological or auditory remediation of specific language impairment (SLI) or language-learning impairment (LLI).

Reference	Age	Participant <i>N</i> (treatment; control)	Impairment or problem	Content of training	Duration of training	Brain research method	Task in testing	Behavioral improvement (pre-test vs. post-test)	Normalization of brain activation
Hayes et al. (2003)	8–12	27 treated; 15 non-treated; 7 non-treated controls	Learning problems, auditory perceptual deficit	Earbics: phonological awareness, auditory processing, language processing	8 weeks	ABR, ERP	Passive listening, attention directed elsewhere	Yes	ERP yes; ABR no
Heim et al. (2013)	6–9	21; 12	LLI	FastForWord*	1 month	EEG	Passive listening and active target detection	Yes	Yes (but not all aspects)
Pihko et al. (2007)	6–7	9 (phonological intervention); 9 (physical exercise)	SLI	Speech and articulation, phoneme discrimination, phonological and linguistic awareness, rapid processing	8 weeks	MEG	Passive listening, attention directed elsewhere	Yes	Yes
Stevens et al. (2008)	6–8	8 treated SLI; 12 treated controls; 13 non-treated controls	SLI	FastForWord*	6 weeks	ERP	Auditory selective attention	Yes	Yes

ABR, auditory brainstem response; EEG, electroencephalography; ERP, event-related potential; MEG, magnetoencephalography. *FastForWord includes auditory discrimination, phoneme identification, phonic match, phonic word, understanding instructions, grammatical structures and rules.

occipito-temporal/occipital dysfunctions, linked with phonological representations and processes (Richlan et al., 2011; Richlan, 2012), seem to be the robustest effects in dyslexia, the effects in the other areas need not to be spurious. The fact that studies use different training techniques and experimental tasks in the scanner during neuroimaging may account for finding remedial changes in different brain functions and areas (Heim et al., 2014).

Neuroimaging research on the effects of auditory and phonological intervention is complemented by ERPs, reflecting the dynamics of neural responses. Studies on dyslexia (**Table 1**) have shown that treatment strengthens brain responses, such as MMN (Kujala et al., 2001; Lovio et al., 2012), attention-related ERP (Stevens et al., 2013), and N400 (Hasko et al., 2014). Lovio et al. (2012) observed also a treatment-induced shortening of the MMN latency across groups receiving grapheme-phoneme or number-knowledge training. In SLI and LLI (**Table 2**), the remedial gains of interventions have been observed as the strengthening of oscillatory brain activity (Heim et al., 2013) and auditory cortical responses, including MMN (Pihko et al., 2007) and attention-related ERPs (Stevens et al., 2013), which are accompanied by improved performance in behavioral language tasks. Training has also been shown to shorten the latency of auditory P1-N2 complex, resulting in a more mature response pattern (Hayes et al., 2003). As the MMN study by Pihko et al. (2007) involved passive listening, where participants' attention was directed elsewhere, enhanced MMN responses indicate remedial effects on low-level, pre-attentive auditory processing that is modified by phonetic representations.

DOES THE CONTENT OF THE INTERVENTION MATTER?

From educators' perspective, it would be important to conduct interventions that tap the core deficit rather than induce compensatory improvements. Direct comparisons using the same experimental tasks but different interventions could clarify, whether the optimal method of remediating language-related deficits can be found. To this end, Shaywitz et al. (2004) have compared the effects of a targeted experimental intervention and a community intervention on behavioral performance and brain activation in children with reading difficulties. The experimental intervention focused specifically on phonological skills with different kinds of tasks, whereas community intervention consisted of activities commonly provided in school, such as remedial reading (see Shaywitz et al., 2004, for details). As a result of interventions, the experimental intervention group had achieved significant gains in reading fluency and showed an increased activation of left-hemisphere brain regions, whereas no such gains were observed after community intervention. This result emphasizes that the nature of intervention is critical for its success (however, see Boot et al., 2013, for discussion on the expectations about improvement).

Besides showing a correspondence between improvements in skills and changes in neural function, neuroscientific measures can illuminate the specific effects of interventions on brain areas subserving distinct cognitive functions. Heim et al. (2014) compared three different kinds of training that focused on phonology, attention, and visual word recognition (reading). They divided school-aged dyslexic children into three training groups according

to their cognitive profiles. All training methods improved children's reading skills to a similar degree. During a reading task in an fMRI scanner, all training programs resulted in the increased activation of the visual word form area, located in the left fusiform gyrus. In some other brain areas, however, the training programs had different effects on brain activation: phonological and reading training increased activation in bilateral parietal areas, whereas attention training increased activation in the left temporal cortex. Thus, different training programs had both shared and specific effects on brain activation, which would not have been evident on the basis of behavior alone.

In line with Heim et al.'s (2014) conclusions on shared effects induced by different training types, very different kinds of interventions have resulted in significant behavioral and neural gains in individuals with language-related deficits. For example, many studies (see **Tables 1** and **2**) have shown the remedial gains of phonological training with FastForWord, including auditory discrimination, phoneme discrimination, phoneme identification, phonic match, phonic word, understanding instructions, and grammatical structures and rules. Significant remedial gains in brain activation and behavior have, however, been obtained also with non-linguistic tasks that, at first sight, might seem to have a less obvious link to language-related deficits. Kujala et al. (2001) presented dyslexics with an intervention with non-linguistic audiovisual training, including matching a sequence of non-speech sounds with a sequence of visual shapes. As a result of intervention, reading accuracy had improved and MMN brain responses to tone-order reversals had increased. The change in reading skills and MMN amplitude significantly correlated, suggesting an association between reading abilities and non-linguistic processing. In a similar vein, Gaab et al. (2007) used non-speech stimuli with rapid transitions to remediate dyslexia. After training, language and reading skills had improved and prefrontal regions associated with the processing of rapid transitions were more strongly activated than before training. The remedial gains for reading skills from very different kinds of intervention tasks allude to the possibility that they tap some common, domain-general process involved in, and perhaps necessary for, reading and language skills and contribute to remedial gains along with domain-specific effects.

A candidate function that may, in concert with others, participate in domain-general remedial gains is attention. Stevens et al. (2008) studied whether linguistic intervention would improve selective attention in children with SLI. As a result of training, measures of receptive language had improved and previously attenuated event-related brain responses reflecting selective attention had normalized. Stevens et al. (2013) have also suggested that children at risk for reading difficulty show atypical brain measures of selective attention, which can be remediated by reading intervention. These findings suggest that language skills and auditory attention are strongly connected, complementing the earlier findings on the role of visual attention in dyslexia (Facioetti et al., 2000; Valdois et al., 2004; Shaywitz and Shaywitz, 2008; Vidyasagar and Pammer, 2010; Franceschini et al., 2012; Vogel et al., 2012). This is in line with models of dyslexia proposing the dysfunction of the inferior parietal lobule, which has

been linked to attention (Richlan, 2012), and the observations of normalized training-induced activation in the inferior parietal areas (Temple et al., 2003; Eden et al., 2004; Meyler et al., 2008).

HOW LONG-LASTING ARE THE REMEDIAL GAINS IN THE BRAIN?

Interventions aim at long-lasting gains. The dynamics of neural changes induced by intervention can be explored with follow-up neuroimaging studies, which enable to specify brain functions that show long-term effects. Shaywitz et al.'s (2004) experimental intervention group of dyslexic children returned to an fMRI scan 1 year after the intervention. The normalization of activation pattern was seen both immediately after intervention as well as 1 year after it, suggesting long-lasting remedial effects. Similarly, Meyler et al. (2008) observed hypoactivation of parietal areas before intervention in poor readers and increased activation of these areas immediately after intervention. Interestingly, when they conducted a follow-up 1 year after the intervention, they found that the activation of the parietal areas had continued to increase. Thus, the activation pattern of previously hypoactive areas had normalized, probably reflecting cumulative learning effects following intervention. These follow-up studies thus show that treatment-induced neurobiological changes, coupled with improvement in behavioral performance, can be long-lasting and may enable cumulative gains in language-related skills.

CONCLUSION

Interventions and training programs involving phonological and auditory tasks have repeatedly gained remedial effects in dyslexia, SLI, and LLI. Neuroscientific research has demonstrated that improved behavioral performance is coupled with changes in both brain function and brain anatomy. Neuroimaging has revealed normalized training-induced brain activation patterns, whereas electrophysiological measures have demonstrated the normalization of strength and timing of brain responses and oscillatory activity after training. Training effects have been observed also in white matter. Especially in the study of dyslexia, neuroscientific studies have illuminated the location of aberrant brain functions, which has enabled to specify the models of the impairment. Neuroimaging studies have also highlighted partly similar and partly specific patterns of neural activation as a result of different training programs. Gains from very different phonological and auditory tasks as well as training effects in the parietal cortex support the models that propose the involvement of some domain-general neural mechanisms, such as attention, in language-related impairments.

In our opinion, neuroscientific studies thus give an important contribution to the treatment of language-related impairments. Specifically, we argue that the use of both neuroscientific and behavioral measures in intervention studies can increase the understanding of how and why interventions change the deficient neural networks, if methodological requirements are met (cf. Bishop, 2013). From educators' perspective, neuroscientific research methods are seldom directly applicable to the assessment of remedial interventions. However, keeping up-to-date in such

research can provide educators with better understanding of the causes of language-related impairments and help them to target interventions more accurately.

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Musical training as an alternative and effective method for neuro-education and neuro-rehabilitation

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In the last decade, important advances in the field of cognitive science, psychology, and neuroscience have largely contributed to improve our knowledge on brain functioning. More recently, a line of research has been developed that aims at using musical training and practice as alternative tools for boosting specific perceptual, motor, cognitive, and emotional skills both in healthy population and in neurologic patients. These findings are of great hope for a better treatment of language-based learning disorders or motor impairment in chronic non-communicative diseases. In the first part of this review, we highlight several studies showing that learning to play a musical instrument can induce substantial neuroplastic changes in cortical and subcortical regions of motor, auditory and speech processing networks in healthy population. In a second part, we provide an overview of the evidence showing that musical training can be an alternative, low-cost and effective method for the treatment of language-based learning impaired populations. We then report results of the few studies showing that training with musical instruments can have positive effects on motor, emotional, and cognitive deficits observed in patients with non-communicable diseases such as stroke or Parkinson Disease. Despite inherent differences between musical training in educational and rehabilitation contexts, these results favor the idea that the structural, multimodal, and emotional properties of musical training can play an important role in developing new, creative and cost-effective intervention programs for education and rehabilitation in the next future.

Keywords: neuro-rehabilitation, neuro-education, music training, music therapy, stroke rehabilitation, language development disorders

Introduction

Recently, the Organisation for Economic Co-operation and Development published the “2012 PISA report” providing strong evidence of the dramatic drop in the scholar level of 15-year-old French pupils over the past 10 years (OECD, 2014). Compared to data collected in 2003, 15 years old French children exhibited a drop of 15 points (from 511 to 495) in mathematics and the number of pupils in difficulty increased dramatically. In addition, learning disorders are very frequently diagnosed during childhood with the prevalence of developmental dyslexia being of 7–10% of the general population (Démonet et al., 2004; Collective expertise INSERM, 2007). These data point

toward the urge of building and testing efficient learning tools to optimize the learning trajectories of typically developing young pupils and even more importantly to remediate specific disabilities found in children with language-based learning impairments. In addition to the burden in the young population, the demographic changes in life expectancy will lead to a significant increase of the population aged over 65 years old in Europe. This population is at high risk of suffering from neurologic age-related diseases (Salomon et al., 2012). For instance, the incidence of stroke is expected to grow by 36% from 2000 to 2025 (Truelsen et al., 2006). The last study of the World Health Organization Global Burden of Disease project is eloquent in showing that stroke remains the second cause of worldwide mortality (Lozano et al., 2012). Although advances in the acute medical management of stroke patients have reduced mortality in high-income countries (Feigin et al., 2014), stroke is still a major cause of disability-adjusted life-years (Murray et al., 2012). Beyond cardiovascular diseases, the rapid aging in Europe will also increase other non-communicable disorders such as neurodegenerative diseases. For instance, Parkinson's disease affects 2.4 per 100 inhabitants of more than 65 years and its prevalence is expected to double by year 2030 (Dorsey et al., 2007). In this context, the need to develop innovative and effective rehabilitation evidence-based techniques is a challenge for the next years in the field of neuro-rehabilitation.

During the last decade, the neuroscientific community has developed a line of research on music perception and on the musician's brain. Results obtained have largely contributed to increase our knowledge on the brain functioning in general and have allowed delineating the positive impact of playing a musical instrument on brain plasticity. In the first part of the review, we report evidence from cross-sectional and longitudinal studies showing that learning to play a musical instrument can induce substantial neuro-plastic changes in cortical and subcortical regions of motor, auditory and speech processing networks. The second part focuses on music to language transfer effect and on the necessary conditions for enhancing language processing in healthy participants. We follow by reporting an overview of the evidence showing that musical training can be an alternative, low-cost, and effective method for the remediation of language-based learning impaired populations. We then focus on neuro-rehabilitation by presenting the results of studies showing that music interventions can enhance motor recovery and neuroplasticity after stroke and can ameliorate motor deficits observed in Parkinson disease. Finally we discuss some of the important similarities and differences between musical training for neuro-education and for rehabilitation purposes.

Musical Practice Fosters Neuroplasticity

In the last decade, the neuroscientific community has concentrated a great amount of effort to explore the positive impact of playing a musical instrument on the brain. Those efforts have provided converging evidence that the musician's brain is an excellent model of neuroplasticity, specifically in the sensory-motor system (Münste et al., 2002; Zatorre, 2013). Indeed, playing in a symphony orchestra requires a large amount of practice: 20-year-old orchestra musicians typically spend more than 10,000 h of musical

practice (Krampe and Ericsson, 1996). During all those hours, the musician will develop and ultimately master many different competences involving sensory-motor, mnemonic, cognitive control, and attentional processes. As a consequence of the repetition of this complex activity, the underlying neural substrates will be eventually modified due to functional and structural neuroplastic mechanisms (see for a recent review, Kolb and Muhammad, 2014).

Neuroplastic Changes in the Sensory-Motor Network

The neuroplastic changes induced by specific training can be studied using both cross-sectional and longitudinal approaches. While longitudinal studies generally use a test-training-retest procedure with naïve participants before training, cross-sectional studies compare a group of experts to a group of laymen participants. In the case of cross-sectional studies in which eventual pre-existing inter-individual differences might account for the differences observed between the groups (Zatorre, 2013) so that causality between music training and the observed effects cannot be demonstrated. By contrast, longitudinal studies with pseudo-random assignment of the participants to a training group and a non-training group allow determining that musical training is the cause of the differences (Schellenberg, 2004). Using a cross-sectional approach, Bangert and Schlaug (2006) conducted a study comparing the anatomical structure of the primary motor cortices in professional musicians and non-musicians. Using magnetic resonance imaging (MRI) these authors compared a group of non-musicians to pianists and violinists. While pianists showed similar structural modifications over both hemispheres, violinists showed a modification only over the right hemisphere. Indeed, playing the piano or the violin involves different type of bimanual control: pianists require fast and accurate finger movements of both hands whereas violinists use both hands asymmetrically favoring fine motor control of left hand fingers and gross motor control of the right hand thus leading to such a structural asymmetry. It is interesting to note that these studies provided evidence on how practicing particular cognitive and motor induce structural plastic brain alterations and improved level of performance for instance in motor areas (Schlaug et al., 1995a; Schlaug, 2001; Schmithorst and Wilke, 2002; Gaser and Schlaug, 2003; Hutchinson et al., 2003; see also Elbert et al., 1995; Draganski et al., 2004; Bengtsson et al., 2005).

Interestingly, the level of musical practice seems to be positively correlated with the increase in gray matter (GM) over motor regions (Gaser and Schlaug, 2003). Nonetheless, a recent study using particularly well controlled and highly selected pianists challenged the results mentioned above by showing a more complex pattern with decreasing GM density in peri-rolandic and striatal areas together with increasing GM density over areas involved in higher order processing such as the right fusiform gyrus, the right mid orbital gyrus, and the left inferior frontal gyrus (James et al., 2014). Increases in GM volume in the putamen have also been associated with timing variability and irregularity of scale playing in professional pianists (Granert et al., 2011). In this study, it was also observed that patients with musical dystonia

presented more volume of GM in the right middle putamen. These surprising results may relate to the age of onset of musical practice and to excessive training, a potentially important factor for influencing plastic mechanisms and neural efficiency (Amunts et al., 1997). For instance, the age of onset of musical training is correlated with the level of motor performance in a rhythm synchronization task (Bailey et al., 2014) and may be decisive for the non-linear dynamic of structural brain changes induced by musical practice (Steele et al., 2013; Groussard et al., 2014).

Musical practice can also modify the strength of the connections between distant areas via white matter modifications. Compared to non-musicians, professional pianists exhibit a larger anterior portion of the Corpus Callosum, the main white matter fiber bundle connecting the two hemispheres (Schlaug et al., 1995b). Using diffusion tractography imaging method (DTI), it has been recently reported that musicians exhibit stronger white matter connectivity in the left and right supplementary motor areas (Li et al., 2014), in the corticospinal tract (Imfeld et al., 2009) and importantly, between auditory and motor areas (Oechslin et al., 2010; Halwani et al., 2011). The strong coupling of motor and auditory networks has been confirmed by further functional imaging studies showing activation of motor areas during the listening to musical rhythms in musicians (Haueisen and Knösche, 2001; Grahn and Brett, 2007; Bengtsson et al., 2009; Grahn and Rowe, 2009). Nevertheless, non-musicians also show such audio-motor co-activation (Baumann et al., 2007).

Moreover, short-term training programs also induce functional plastic changes. When beginners learn to play simple melodies (Bangert and Altenmüller, 2003). For instance, Lahav et al. (2007) trained non-musicians to play melodies by ear during five consecutive days and found activations in premotor regions when participants passively listened to the trained melodies (see also Meister et al., 2005). By contrast, Chen et al. (2012) observed a reduction of activation in both dorsal and ventral premotor cortices during musical training of naïve participants. This pattern of reduced activation over motor areas in musicians may reflect functional efficiency changes induced by musical training (Jäncke et al., 2000). In this context, Pascual-Leone et al. (1995) were the first in observing functional reorganization during piano learning using transcranial magnetic stimulation (TSM). After 4 weeks of training, participants showed a reduction of the motor map while showed an increase during the first week. This reorganization process over cortical motor maps highlight three important aspects. Firstly, rapid functional changes can occur after a rather short period of training. Secondly, the plastic changes induced by short-term training disappear when the training ends (see for example also Draganski et al., 2004). Thirdly, long-term training can lead to more efficient reduced patterns of activation. Aside from motor regions, plastic changes have also been reported at the level of the somatosensory cortices: musicians are more sensitive to tactile stimulations of the fingers than non-musicians, suggesting that musical practice can modify the size of the somatosensory receptive fields (Elbert et al., 1995).

Playing a musical instrument requires a clear motor component for a good performance. Nonetheless, the auditory dimension is also crucial in order to generate error feedback that might correct or adjust movements in case of errors (Maidhof, 2013; Maidhof

et al., 2013; Pfordresher and Beasley, 2014) and to accurately perceive the pertinent acoustical parameters of the auditory input. Therefore, music induced plastic modifications over auditory areas might also be expected to occur.

Neuroplastic Changes in the Auditory Pathway

The positive effects of musical practice on auditory processing have been evidenced by several studies showing lower discrimination thresholds for frequency, duration, silences, or time intervals in experts than in laymen (Jones et al., 1995; Kishon-Rabin et al., 2001; Micheyl et al., 2006; Rammsayer and Altenmüller, 2006; Mishra et al., 2014). In terms of structural plasticity, as in the case of the motor system, musical practice induces structural changes in the cortical auditory network: GM changes have been observed in both longitudinal and cross-sectional studies with musicians exhibiting enlarged AC compared to non-musicians (Schlaug et al., 1995a; Keenan et al., 2001; Schneider et al., 2002, 2005; Bermudez and Zatorre, 2005; Hyde et al., 2009). Moreover, compared to non-musicians, musicians also show more developed superior longitudinal fasciculus and arcuate fasciculus, the fiber bundles connecting the AC to Broca's area (Oechslin et al., 2010; Wan and Schlaug, 2010; Halwani et al., 2011).

In line with these findings, functional changes have been reported in musicians at almost every single step of the auditory pathways: from the cochlea to the inferior colliculus (IC) and finally to the auditory cortices (AC). Musicians show functional changes already at the very peripheral level with enhanced activity of the Medial olivocochlear complex, responsible for controlling the cochlear micromechanics (Perrot et al., 1999; see for a review Perrot and Collet, 2014). Since almost 10 years, the systematic work of Nina Kraus and her group have provided exciting evidence that musical practice also induces substantial neuroplastic changes over subcortical structures. The IC, a subcortical structure in the brainstem receiving both bottom-up inputs from the cochlea and top-down inputs via the corticofugal pathway (Kraus and Chandrasekaran, 2010), encodes specific characteristics of the auditory input (Russo et al., 2004). For instance, very brief acoustic events such as stop consonants ("b," "p," "g," "d," "t") are reflected by the transient response of the auditory brainstem responses (ABRs) whereas sustained acoustic events such as vowels are reflected by the frequency following response (FFR). Compared to non-musicians, adult musicians show more robust transient response and FFR to both musical and speech sounds (Musacchia et al., 2007; Parbery-Clark et al., 2012) suggesting that at the level of the brainstem, the representations of the sounds are more elaborated and more accurate in musicians than in non-musicians. The enhancement of ABRs is already visible at age three suggesting that very few years of musical training might be sufficient to elicit such consistent plastic changes in the IC (Strait et al., 2013). Moreover, the benefit of musical training received during childhood appears to remain during adulthood as revealed by a correlation between ABR amplitude and how recently participants quitted the training (Skoe and Kraus, 2012; see also Strait and Kraus, 2011).

Finally, musical practice fosters functional brain plasticity in cortical areas: compared to non-musicians, adult and child musicians show enhanced response of the AC as reflected by larger N1/P2 amplitude to complex sounds (Shahin et al., 2003,

2004; Trainor et al., 2003). Moreover, this enhanced N1/P2 is particularly visible when the stimulus presented is the instrument played by the participants (Pantev et al., 1998, 2001). Again, these results suggest that musicians have a more elaborated representation of the auditory input and better encode fine-grained features of sounds than non-musically trained individuals.

Interestingly, musical practice also increases the neural sensitivity to the statistical regularities found in the auditory input (François and Schön, 2014). For instance, a deviant sound rarely occurring within a sequence of repeated standard sounds elicit a specific event-related potential (ERP) component, the mismatch negativity (MMN), reflecting the pre-attentive detection of an auditory change (Näätänen et al., 2005; Grimm and Escera, 2011). Adult and child musicians often show larger and/or earlier MMNs to changes in several features of the sounds such as frequency, duration, intensity, or spatial localization than non-musicians (Tervaniemi et al., 2001, 2005; Van Zuijen et al., 2005; Vuust et al., 2005, 2011; Brattico et al., 2009; Marie et al., 2011; Putkinen et al., 2014). Additional evidence shows that musicians exhibit enhanced MMN to change in longer sequences of structured sounds (Tervaniemi et al., 2001; Trainor et al., 2002; Fujioka et al., 2004; Habermeyer et al., 2009).

Neuro-Education: Music Training as an Alternative Tool to Promote Literacy Skills

After having delineated the positive effects of playing music on brain plasticity in the auditory and motor networks, we now turn to the topic of neuro-education. We aim to give an overview of evidence showing that language-based learning impaired children often show deficits in the specific processes that are boosted by musical practice. We then present the hypothesis of music to language transfer effect, which allow grounding the subsequent evidence from both cross-sectional and longitudinal studies showing the beneficial effects of musical practice on speech processing in typically developing children. We finally summarize the few studies testing music-training programs in language-based learning impaired populations.

Speech Processing in Children with Language Impairment

Children with language based learning impairments such as developmental dyslexia present a specific deficit in reading despite conventional instruction, socio-cultural level, normal intelligence and the absence of sensory deficits (Lyon et al., 2003; Vellutino et al., 2004; Collective expertise INSERM, 2007). Phonological awareness is crucial for normal language development (Ramus, 2003; Serniclaes et al., 2004) and relies on the ability to categorize speech sounds on the basis of extremely short timing differences. The voice onset time (VOT) is an acoustic parameter largely used to study phonological awareness as it allows differentiating the sound “ba” from the sound “pa,” a task in which dyslexic children have difficulties (Serniclaes et al., 2004). In line with these findings, Chobert et al. (2012) were able to demonstrate that children with dyslexia are impaired in the pre-attentive processing of VOT and duration in syllables based on MMN data. Children with

language learning difficulties have also difficulties in extracting speech sounds when presented in a background noise (Ziegler et al., 2005, 2009) and show degraded neural responses to speech in noise stimuli (Warrier et al., 2004; Wible et al., 2004; Anderson et al., 2010).

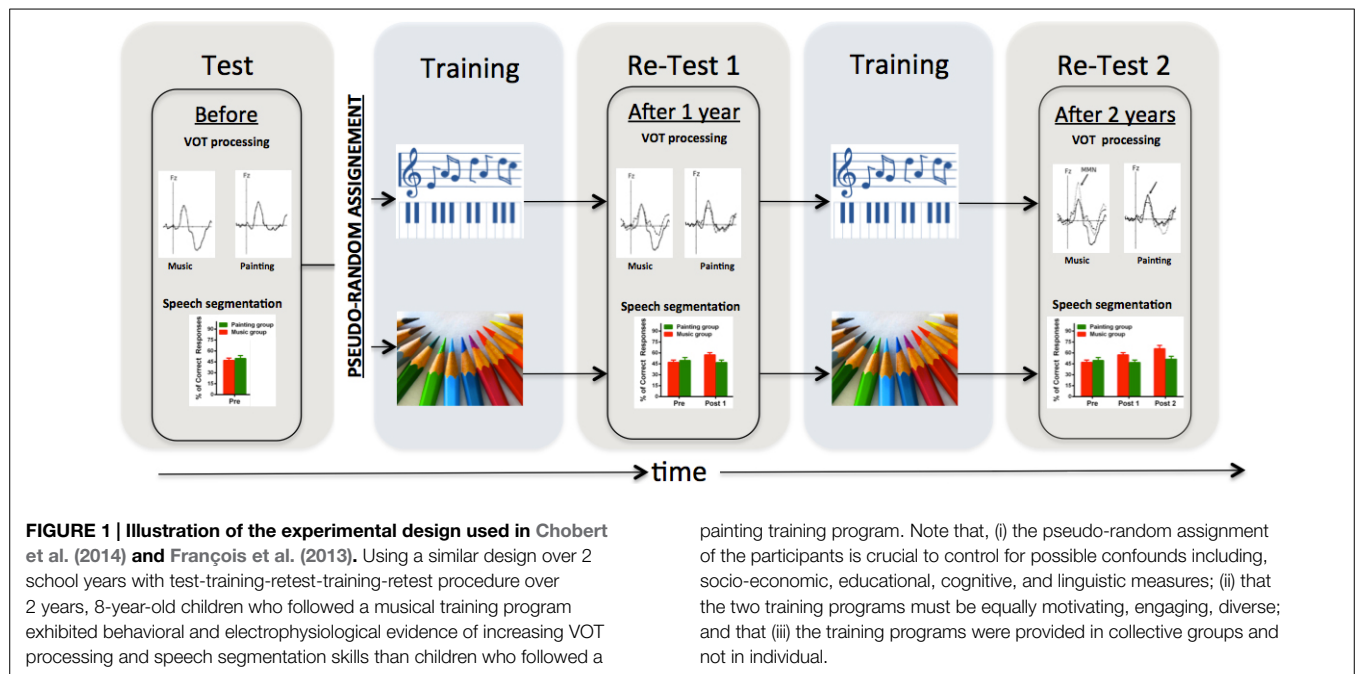
These children might present a general deficit in the processing of timing information (Goswami et al., 2011). This timing hypothesis would explain why they exhibit impaired phonological processing (Ziegler and Goswami, 2005) and impaired general rhythmic processing (Thomson and Goswami, 2008; Corvieu and Goswami, 2009). Huss et al. (2011) showed that children with dyslexia present a deficit in the perception of rise time, an acoustic parameter important to extract the periodic and eventually metrical structures of speech (Cummins and Port, 1998; Quené and Port, 2005; Goswami, 2010; Huss et al., 2011). Growing evidence converge on the idea that rhythmic skills are crucial for the development of literacy skills in typically developing children (Tierney and Kraus, 2013; Woodruff Carr et al., 2014) and that children with dyslexia are impaired at tapping to a rhythm, and in perceiving tempo (Thomson and Goswami, 2008; Corvieu and Goswami, 2009). These results also confirm findings showing a link between literacy skills, phonological abilities, and musical aptitudes in typically developing child and in adult participants (Anvari et al., 2002; Slevc and Miyake, 2006; Tierney and Kraus, 2013). Following this line, a recent study with 48 children with dyslexia shows that temporal auditory processing strongly predicts phonological processing and reading abilities (Flaunagacco et al., 2014).

Music to Language Transfer of Competences: Why and How Music Can Transfer to Language

Transfer of skills generally occurs when a specific skill acquired in one specific domain influences processes in another supposedly unrelated domain. Several observations have lead to the hypothesis that musical practice could transfer to language and more specifically to speech processing (Kraus and Chandrasekaran, 2010; Besson et al., 2011; Patel, 2011, 2014). Firstly, as presented above, there is a whole body of literature showing enhanced auditory processing in musicians. Secondly, music and speech share similarities: both involve the processing of similar acoustic cues (such as pitch, intensity, timbre, and duration) and both involve maintaining sequences of sounds that are unfolding in time in a structured manner. Thirdly, music and speech processing show a clear overlap in their cortical and subcortical neural substrates (Koelsch et al., 2005; Vigneau et al., 2006; Schön et al., 2010), suggesting shared neural resources (Patel, 2011, 2014).

Music to Language Transfer Effects, Evidence in Healthy Children

In the specific context of education, it is now clearly demonstrated that noisy environments in classroom settings with higher than normal ranges level of noise negatively impacts pupils' performance (Knecht et al., 2002; Shield and Dockrell, 2008). Indeed, the level of performance in a syllable discrimination task dramatically drops as the level of noise increases (Neuman et al., 2010). The amplitude and latency of ABRs are also clearly reduced in the



presence of acoustic noise (Burkard and Sims, 2002; Russo et al., 2004). Children with language based learning impairment show impaired speech in noise perception and altered neural responses to speech sounds presented in a background noise. Musical practice seems to be a good tool to prevent this deleterious effect of noise as adult musicians exhibit more preserved ABRs in noise than non-musicians (Parbery-Clark et al., 2009a,b; Bidelman and Krishnan, 2010; Strait et al., 2012). Furthermore, a recent longitudinal study has revealed that musical training in high school music classes can induce these changes (Tierney et al., 2013), which appear to maintain during lifespan (Zendel and Alain, 2012, 2013).

If musicians are better in perceiving speech in noise they also have refined representations of syllables (Degé and Schwarzer, 2011; Zuk et al., 2013; see also Moreno et al., 2009) at both subcortical and cortical levels (Chobert et al., 2011, 2014; Strait et al., 2012; see **Figure 1** for an illustration of the experimental design). All together, these results show that musicians have better neural encoding of speech sounds, which might help to develop a greater sensitivity to the metrical structure of speech (Port, 2003). Compared to non-musicians, adult and child musicians showed increased sensitivity to subtle pitch modifications inserted in the prosodic contour of sentences being uttered in their native or in a foreign language (Schön et al., 2004; Magne et al., 2006; Marques et al., 2007). Adult musicians are also more sensitive to the metrical structures of speech and to anomalous durational modifications in sentences (Marie et al., 2011).

Further evidence of better speech processing skills in musicians than in non-musicians was also provided by cross-sectional and longitudinal studies exploring speech segmentation ability in adults and children (François and Schön, 2011; François et al., 2013, 2014). Speech segmentation is one of the mandatory steps for acquiring a new language, which requires the ability to extract words from continuous speech. When presented with 2 min of

an artificial stream of statistically structured syllables, infants and adults are able to segment and discriminate syllables sequences that are part of the stream (i.e., familiar sequences) from new sequences (i.e., unfamiliar sequences; Saffran et al., 1996; Aslin et al., 1998). While adult musicians barely outperformed non-musicians (François and Schön, 2011), musically trained children largely outperformed their non-musician counterparts after 1 year and even more after 2 years of musical training (François et al., 2013). Moreover, neural responses in both adult and child musicians differentiated familiar from unfamiliar sequences during the behavioral test. In a further study, François et al. (2014) provided evidence that the ability to differentiate familiar from unfamiliar items was correlated with how fast a fronto-central negative ERP component emerged during the exposition to the artificial speech stream.

The mounting evidence of the beneficial effects of musical training on speech auditory processing in typically developing children as well as the timing deficits found in children with language based learning impairments led researchers to test the idea that musical training could be used as a remediation tool.

Music Training Programs in Language Impaired Populations

Overy (2000, 2003) were the first evaluating the efficiency of playing music in children with dyslexia. Despite a small group and no clear matched-control, the results showed improved phonological awareness and spelling performance after a rhythmic training program. Along these lines, short rhythmic priming sequences have been used to enhance phonological processing in typically developing children and in prelingually deaf children (Cason and Schön, 2012; Cason et al., 2015) and to improve syntactic processing in children with language impairments (Przybylski et al., 2013). Despite a clear lack of controlled trials in children and adolescents with dyslexia (Cogo-Moreira et al., 2012), a recently published

study confirms that musical training can improve reading skills and educational achievement in those children (Cogo-Moreira et al., 2013). Moreover, a recent study from Bishop-Liebler et al. (2014) provides clear evidence of the positive influence of musical practice on the timing deficits found in dyslexics.

Finally, recent data suggest that an adapted musical training program can enhance auditory, phonological, and cognitive processes in 8-year-old deaf children (Rochette et al., 2014) and that musical rhythmic priming can enhance phonological production in prelingually deaf children (Cason et al., 2015). These results are important for future studies in cochlear-implanted users who show profound deficits in speech in noise perception, in the perception of complex rhythms, timbres, and melodies with somewhat preserved tempo and simple rhythms perception (McDermott, 2004; Drennan and Rubinstein, 2008).

Neuro-Rehabilitation: Music-Based Therapies in Neurologic Population

Music playing requires the processing of multimodal information and entails high neural demands. Multimodality is an excellent opportunity to adapt different musical activities such as playing an instrument, moving in synchrony along a rhythm or listening to music with a therapeutic purpose (Pantev and Herholz, 2011). In this section, we present rehabilitative techniques using music as a key feature to remediate motor deficits in neurological conditions.

Playing Music to Overcome Motor Deficits in Stroke

Motor impairment after stroke refers to a weakness of the muscles mainly affecting the control and performance of voluntary movements (Mohr et al., 2011). This deficit is the most common outcome after stroke with paresis of the upper and lower extremities being found in almost 70% of cases (Rathore et al., 2002). While paresis of the legs impedes functional mobility such as walking, the limitations of arm and hand paresis extend to several daily-living activities (Langhorne et al., 2011).

Music-supported therapy (MST) aims to restore paresis of the upper limb through musical instrument playing (Schneider et al., 2007; see **Table 1**). In order to enhance fine and gross movements, patients are trained on playing melodies on a midi piano and/or electronic drum pads. MST relies on four basic principles: massive repetition, audio-motor coupling, shaping, and emotion-motivation effects (Rodríguez-Fornells et al., 2012). Firstly, MST requires massive repetition of simple sequences of movements through the intervention. Importantly, high-intensity practice is a basic and well-accepted principle in neuro-rehabilitation (Langhorne et al., 2009, 2011). Secondly, multimodal integration may enhance audio-motor coupling where the musical sound serves as a feedback to reinforce the movement, to correct the errors, to adjust the timing and to refine motor representations. Thirdly, the therapy is adapted to the level of impairment and to the progression of the patient. Fourthly, the emotional-motivational aspects of music may regulate emotional responses through the playfulness of learning a new skill.

Music-supported therapy is successful in reducing the motor deficits in subacute stroke patients. Altenmüller et al. (2009) and

Schneider et al. (2007) compared the effectiveness of MST to conventional treatment. Only patients in the MST group improved in frequency, velocity and smoothness of fingers and hand tapping movements. Moreover, those patients obtained greater scores over time on standardized clinical tests assessing motor functions. However, to what extent is the presence of music responsible for the observed gains and associated plasticity? In a single-case study (Rojo et al., 2011), a patient performed a passive listening task with unfamiliar and trained melodies. Interestingly, while before the application of MST the patient exhibited only activation of the AC, the motor regions were also activated after the training. This phenomenon of audio-motor coupling provided evidence that the auditory feedback is an essential part of the therapy by contributing to enhance activations over motor regions (Rodríguez-Fornells et al., 2012). In a further study, Amengual et al. (2013) used TMS to demonstrate changes in the excitability of the sensorimotor cortex due to MST. Participants recovered from their motor deficits and exhibited an increased excitability of the sensorimotor cortex in the affected hemisphere after 4 weeks of MST. Moreover, a lateral shift in the motor map of chronic patients was evidenced after the training and was associated with motor gains. Interestingly, similar results have been recently reported in subacute patients (Grau-Sánchez et al., 2013). Taken together these studies suggest that MST can induce functional changes associated to brain reorganization processes.

Recent studies aimed at modifying different aspects of the MST protocol. Van Vugt et al. (2014) implemented MST in two groups of subacute stroke patients in which participants received the therapy in pairs instead of individual sessions. One group had to play together while the other group played in turns. Although both groups improved their performance in a motor task, results indicated a positive trend favoring the in-turn group. Besides, the in-turn group improved more their mood as well as their feelings about their partner. The idea that music playing is a shared experience (Overy, 2012) is interesting to consider because patients can feel emphatic and understood by others individuals presenting similar difficulties. This could in turn enhance the mood and reduce depressive symptoms (Gillen, 2010). MST has also been evaluated in a home-training protocol (Villeneuve et al., 2014) showing improvements in the paresis of chronic patients that were maintained over time. During nine 1-h sessions, patients played musical sequences using Synthesia (Synthesia LLC) a software that provides visual cues to guide them. The sessions were complemented with at home exercises on a roll-up piano. Home sessions in chronic stages may be an appropriate cost-effective approach once patients have gained a certain degree of improvements and stabilization. In this vein, the use of new technologies and adapted software with rehabilitative purposes opens new directions in the field of neuro-rehabilitation. The recently developed MusicGlove (Friedman et al., 2014) may be an alternative tool to treat paresis in chronic stages. The MusicGlove is an instrumented glove that produces notes during gripping movements while being guided with visual stimuli displayed on a screen. An exploratory study with 12 chronic patients has revealed improvements in motor functions compared to conventional therapy or isometric training (Friedman et al., 2014).

TABLE 1 | Summary of the studies evaluating MST to restore upper limb paresis in stroke patients.

Study	Participants	MST program	Results
Schneider et al. (2007)	Subacute patients MST group (<i>n</i> = 20) CT group (<i>n</i> = 20)	15 sessions of 30 min during 3 weeks Piano and drum playing	MST group: increased frequency, velocity, and smoothness in a finger and a hand-tapping task. Improvements in ARAT, BBT, 9HPT, APS motor test CT group: no improvements
Altenmüller et al. (2009)	Subacute patients MST group (<i>n</i> = 32) CT group (<i>n</i> = 30)	15 sessions of 30 min during 3 weeks Piano and drum playing	MST group: increased frequency, velocity, and smoothness in a finger and a hand-tapping task. Increased smoothness in prono-supination movements and velocity in reaching a target. Better scores in ARAT, BBT, 9HPT, and APS motor test CT group: no improvements
Rojo et al. (2011)	Chronic patient Case study	20 sessions of 30 min during 4 weeks Piano and drum playing	Increased smoothness in a finger and a hand-tapping task and in prono-supination movements. Increased frequency in a hand-tapping task. Increased amplitude of motor-evoked potentials in both hemispheres. Reduced neural activation in the unaffected hemisphere during a motor task with the paretic hand. Functional activation of motor regions during the passive listening of trained sequences
Amengual et al. (2013)	Chronic patients MST group (<i>n</i> = 20) Healthy group (<i>n</i> = 20)	20 sessions of 30 min during 4 weeks Piano and drum playing	Increased frequency in a finger-tapping task, increased smoothness in a hand-tapping task. Better scores in ARAT motor test. A lateral shift in the representational motor cortical map. Increased amplitude of motor-evoked potentials in the affected hemisphere Healthy group: no improvements
Grau-Sánchez et al. (2013)	Subacute patients MST group (<i>n</i> = 9) Healthy group (<i>n</i> = 9)	20 sessions of 30 min during 4 weeks Piano and drum playing	Improvements in ARAT, BBT, and APS. Increased quality of life. Increased excitability in the affected hemisphere and a posterior shift in the representational motor cortical map Healthy group: reduction in the area of the representational motor cortical map
Van Vugt et al. (2014)	Subacute patients MST in turn group (<i>n</i> = 14) MST together group (<i>n</i> = 14)	Three individual sessions and seven sessions in pairs, where one group played in turns with their couple and the other group played in synchrony with their couple. In total, 10 sessions of 30 min over the course of 3 or 4 weeks Piano playing	Both groups improved in 9HPT test, but the in turn group improved more. More synchrony in a index-to-thumb tapping in both groups. Reduction in depression and fatigue in both groups. Both improved mood but the in-turn group became more positive over the therapy. The in-turn group rated higher how they experienced sessions and how they felt with partner
Villeneuve et al. (2014)	Chronic patients MST group (<i>n</i> = 13) No control group but intrasubject design	Nine individual sessions of 1 h guided by a therapist and six sessions of 30 min at home without therapist. In total, 15 sessions 3 weeks Piano playing	Better scores in BBT, 9HPT, FTN, FTT, and Jebsen motor test. Improvements maintained after 3 weeks of finishing the treatment

The abbreviations in the participants column correspond to: MST, music-supported therapy; CT, conventional treatment. The abbreviations in the results' column refers to the following motor tests: ARAT, Action Research Arm Test (Carroll, 1965; Lyle, 1981); BBT, Box and Blocks Test (Mathiowetz et al., 1985); 9HPT, 9 Hole Pegboard Test (Parker et al., 1986); APS, arm paresis score (Wade et al., 1983); FTN, Finger To Nose Test; FTT, Finger Tapping Test; Jebsen, Jebsen Hand Function Test (Jebsen et al., 1969).

Importantly, all subacute patients involved in these studies receive a rehabilitation program in a hospital or outpatient setting that includes physiotherapy and occupational therapy to train the affected extremity (Gillen, 2010). This may limit the interpretation of the positive effects of MST over conventional treatments because participants cannot be excluded from the standard rehabilitation program (Oremus et al., 2012). Moreover, natural brain processes of recovery take place in acute and subacute stages which may be a confounding factor (Cramer et al., 2011; Johansson, 2011; Zeiler and Krakauer, 2013). In order to control for spontaneous recovery, it is important to have comparable groups in terms of age, severity of the deficits and time since stroke. Chronic stages are characterized by a stabilization of the deficits via compensatory mechanisms at both the behavioral and neural levels (Cramer et al., 2011; Langhorne et al., 2011) and thus, it might be more appropriate to perform proper randomized controlled trials (RCT) using

within-participant designs combined together with disease progression models.

Using Music Listening to Improve Gait in Parkinson's Disease and Other Neurological Diseases

The main symptom in Parkinson's disease is motor impairment in gait, which is characterized by a decreased speed, shorter stride length, and asymmetries in stride times for both lower limbs, all in turn increasing cadence of steps. This pronounced reduction in speed and amplitude is accompanied by a difficulty in initiating voluntary movements in later stages of the disease, with patients experiencing a freezing of the movements (Okuma and Yanagisawa, 2008). This limitation in walking will in turn impair balance and postural control and lead patients to a reduced activity and high risk of falls (Kim et al., 2013). A dysfunction of the basal

ganglia is responsible for the mentioned symptoms (Stoessl et al., 2014) and the efficacy of pharmacological treatment reduces with time. Thus, the development of behavioral therapies may help in coping with the impairment and may be an alternative to be combined with pharmacological therapy.

One approach to enhance intrinsically rhythmical movements consists in using external sensory cues to entrain the movements (Thaut and Abiru, 2010; Thaut et al., 2014). Rhythm auditory stimulation (RAS, Thaut et al., 1996) aims to facilitate gait using metronome beats. The patient is first trained to move to the beat and then the tempo is increased from 5 to 10% over the baseline to accomplish faster movements. A first study observed that 3 weeks of RAS could improve gait velocity, stride length, and step cadence more than no treatment or self-paced training program (Thaut et al., 1996). Further studies have confirmed the positive effects of RAS on gait parameters such as overcoming freezing of gait (McIntosh et al., 1997, 1998; Thaut et al., 1997; Freedland et al., 2002; Fernandez del Olmo and Cudeiro, 2003, 2005; Arias and Cudeiro, 2010; for a review, see Nombela et al., 2013). It also improves stride length, gait velocity, cadence, and asymmetry in patients with stroke (Thaut et al., 1997, 2007; for a review in other neurological conditions, see Wittner et al., 2013). The neuroplastic mechanisms beyond the effectiveness of RAS may be due to an increased activity in the cerebellum, trying to compensate the dysfunctional pathway of the basal ganglia to regions of the pre-motor cortex (Fernandez del Olmo and Cudeiro, 2003). However, two studies have showed more benefits in the advanced than in the early stages of the disease suggesting that the effectiveness of RAS depends on the stage of the disease (Willems et al., 2006; Arias and Cudeiro, 2008). Moreover, some studies have explored variations in RAS, manipulating the tempo (Fernandez del Olmo and Cudeiro, 2003, 2005) as well as the rhythm with respect to the individual's baseline (Willems et al., 2006; Ledger et al., 2008). These studies have evidenced that beats presented at 20% slower than the baseline cadence does not benefit gait (Willems et al., 2006). Although other types of therapy have examined the use of other sensorial modalities (visual and proprioceptive cues), the auditory modality seems to be the best to improve gait in Parkinson's disease (Nombela et al., 2013).

Music Therapy and Emotion in Neuro-Rehabilitation

Beyond motor impairment, neurological patients are at high risk for suffering psychological consequences. Around one third of stroke patients suffer from depression in the following months and years (Hackett et al., 2005; Ayerbe et al., 2013) and apathy and anxiety can also be found as a frequent neuropsychiatric consequence (Campbell Burton et al., 2011; Caeiro et al., 2013). These neuropsychiatric symptoms are thought to impact health-related quality of life, increase morbidity and worsen the cognitive impairments (Whyte and Mulsant, 2002; Aarsland et al., 2012; Ayerbe et al., 2013). Psychological factors can also have a negative effect on recovery and can affect the engagement in the rehabilitation program. Pharmacological interventions have small effects on treating depression and reducing its symptoms in stroke and Parkinson's disease and can also lead to negative side effects (Hackett et al., 2008, 2010; Aarsland et al., 2012).

Some studies have reported that MST can reduce depression and fatigue and can improve the quality of life in stroke patients (Grau-Sánchez et al., 2013; Van Vugt et al., 2014). Music playing could be an alternative approach to target depression and neuropsychiatric symptoms through emotion regulation. However, there is little research studying the effectiveness of music therapy in the emotional domain. Compared to listening to audio books or auditory intervention, the daily listening to self-selected music during 2 months improves mood in the following months. Listening to music can also improve verbal memory and focused attention (Särkämö et al., 2008). Importantly, listening to music also induced structural changes with an increase of GM in frontal and limbic structures (Särkämö et al., 2014). Participants reported that music was helpful for relaxing and sleeping, influenced their mood and evoked memories and reflexive thoughts (Forsblom et al., 2009). Musical activities may be a tool to modulate the emotional reactions and cope with them through playfulness activity.

Future Research in the Field of Music Therapy in Neuro-Rehabilitation

We focused our review on MST, RAS, and listening to music as standardized therapies to treat motor deficits and improve mood. Further research should aim to standardize interventions to build a strong dataset in this field. Moreover, the majority of studies refer to conventional treatment as the current rehabilitation program provided by the hospital. However, the content of the rehabilitation program may vary depending upon the facilities or the countries. Thus, an accurate description of the exercises performed by the control group is needed. Randomization of participants and blind evaluations of the treatment constitute RCTs and are necessary to implement interventional programs in clinical practice.

Musical Training for Neuro-Education and Neuro-Rehabilitation: Differences and Similarities in Conceptual and Practical Aspects

Fundamental differences may exist between musical training for education or rehabilitation. Firstly, conceptual differences on the aim of musical training remain in the two cases. In neuro-education, musical training generally aims to boost a "typical" developmental trajectory (Kraus and Chandrasekaran, 2010), whereas in neuro-rehabilitation, musical training is rather used to normalize or compensate sensory, motor or cognitive deficits induced by a pathological condition (Cramer et al., 2011). Nonetheless, in the case of children with dyslexia, musical training will also aim to normalize the learning trajectories in order to facilitate speech and language processing which in turn may have a positive impact on educational achievement (Tierney and Kraus, 2013).

Secondly, the neuro-plastic mechanisms induced by musical training during childhood or in neurological population may be different. This might be due to the intrinsic physiological differences at play in these two cases. Animal and human studies

showed that auditory stimulation received early in life enhances both sub-cortical and cortical electrophysiological responses to sounds (Sanes and Constantine-Paton, 1985; Kraus and Chandrasekaran, 2010) and persist in adult development (Zhang et al., 2001; Skoe and Kraus, 2012; White-Schwoch et al., 2013). Early in life, the neural system is immature and capitalizes on plastic changes such as myelination, neurogenesis, or dendritic growth (Kolb and Telskey, 2012; Kolb et al., 2012). On the contrary, stroke generally occurs in a mature neural system in which the insult has induced the death of neurons from a specific region and the disruption of neural networks. The plastic mechanisms underpinning recovery after stroke rely on different processes than in typical development such as restitution and substitution (Albert and Kesselring, 2012). Studies exploring the benefits of MST have shown that musical training induced functional reorganization of cortical motor maps (Amengual et al., 2013). In Parkinson disease, the aim of the therapy is to compensate the deficits in internally generated movements using music as an external cue that engages a different motor pathway to achieve the same goal, which is the initiation and normalization of gait (Nombela et al., 2013).

Thirdly, qualitative and quantitative practical differences in the type of training administered also remain. In neuro-rehabilitation settings, the training is generally provided during a relatively short period of time together with a high intensity (Albert and Kesselring, 2012). Interestingly, the use of the affected upper extremity during sessions of conventional stroke rehabilitation is minimal (Birkenmeier et al., 2010; Krakauer et al., 2012), suggesting that rehabilitation protocols should increase the doses of practice. In this context, musical training protocols for motor rehabilitation may be a good choice as they involve the massive repetition of movements with a high-intensity. Moreover, MST and RAS are most of the time individually administered and the complexity of the tasks is adapted to the progression of each patient. In the case of MST and particularly in middle to low-income countries, the training may become rather expensive due to the individualized administration of the therapy. However and importantly, when patients are discharged from the rehabilitation unit, they are most of the time physically inactive, exhibiting sedentary behaviors and have poor social interactions (Särkämö et al., 2008; Dontje et al., 2013; Tieges et al., 2015). The use home-based MST and RAS therapies may be important to encourage these patients to continue the rehabilitation and to maintain active. Moreover, listening to music could be good alternative not only to be applied at home but also in the rehabilitation unit, where most of the time patients remain in their rooms with no social interaction (Särkämö et al., 2008). Music making in neuro-education settings is generally provided in group settings with a lower intensity than in neuro-rehabilitation but importantly, the training is administered during longer periods of time thus allowing reaching a higher degree of musical complexity than in the context of rehabilitation. The group settings used with children may be more cognitively demanding than the ones used with neurologic patients. Most of the time, children have to play in synchrony with each other and ultimately create new musical pieces whereas patients will generally listen and reproduce simple familiar musical pieces.

Despite obvious differences, fundamental similarities might relate to the emotional, sensory, motor, cognitive, and social

demands of music making *per se* (Herholz and Zatorre, 2012). In both fields, musical training is selected for its unique characteristics involving complex interactions between different domains and systems. Due to its multimodal aspect, musical training represents a good activity to develop audio-motor interactions, for cognitive stimulation and mood regulation. Moreover, music making as well as music listening are generally pleasant activities that are most likely to induce motivated behaviors. This is particularly the case for patients who may be more committed toward an enjoyable activity with a specific purpose rather than to a repetitive training with different rehabilitative tools. Musical training is also able to reinforce social cohesion or bonding through repetitive inter-individual interactions (Huron, 2001). Another similarity may also reside on the fact that musical practice will induce positive side effects: by enhancing language processing for the educational side and by boosting spared functions for the rehabilitation side. Finally, the permanence in time of the benefits of music making is clearly observed and is probably the most meaningful aspect for both purposes.

Conclusion

The neural mechanisms of music-induced plasticity are still not perfectly understood (Fukui and Toyoshima, 2008), but the evidence for a positive effect of musical practice are growing and could justify the use of music both in the context of neuro-education (Caine and Caine, 1990) and of neuro-rehabilitation (Särkämö et al., 2014). The findings showing that the age of onset of musical training influences the dynamic of training induced plastic changes (Steele et al., 2013; Groussard et al., 2014) leads to the idea that multiple sensitive periods for specific functions and specific brain networks may co-exist in typical development (Penhune, 2011). This opens interesting perspectives to study the benefit of musical training in the developing brain as well as to study its consequences on speech perception and scholar achievement. The recent findings showing that listening to music is a rewarding experience for most of the people (Mas-Herrero et al., 2014) and that simple music listening activates the rewarding dopaminergic system (Salimpoor et al., 2013) give even more support to the idea that musical practice may be the perfect tool for neuro-education and Rehabilitation by fostering plastic changes in the healthy or pathological brains. Despite these growing evidence, the educational and health systems generally seem to be refractory to the idea of developing musical training programs. We hope that both teachers and therapists will keep on believing and applying alternative methods based on musical practice.

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Music and Dyslexia: A New Musical Training Method to Improve Reading and Related Disorders

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Numerous arguments in the recent neuroscientific literature support the use of musical training as a therapeutic tool among the arsenal already available to therapists and educators for treating children with dyslexia. In the present study, we tested the efficacy of a specially-designed Cognitivo-Musical Training (CMT) method based upon three principles: (1) music-language analogies: training dyslexics with music could contribute to improve brain circuits which are common to music and language processes; (2) the temporal and rhythmic features of music, which could exert a positive effect on the multiple dimensions of the “temporal deficit” characteristic of some types of dyslexia; and (3) cross-modal integration, based on converging evidence of impaired connectivity between brain regions in dyslexia and related disorders. Accordingly, we developed a series of musical exercises involving jointly and simultaneously sensory (visual, auditory, somatosensory) and motor systems, with special emphasis on rhythmic perception and production in addition to intensive training of various features of the musical auditory signal. Two separate studies were carried out, one in which dyslexic children received intensive musical exercises concentrated over 18 h during 3 consecutive days, and the other in which the 18 h of musical training were spread over 6 weeks. Both studies showed significant improvements in some untrained, linguistic and non-linguistic variables. The first one yielded significant improvement in categorical perception and auditory perception of temporal components of speech. The second study revealed additional improvements in auditory attention, phonological awareness (syllable fusion), reading abilities, and repetition of pseudo-words. Importantly, most improvements persisted after an untrained period of 6 weeks. These results provide new additional arguments for using music as part of systematic therapeutic and instructional practice for dyslexic children.

Keywords: dyslexia, music therapy, phonology, reading, attention, learning disorders

INTRODUCTION

There is worldwide agreement for estimating between 5 and 15% the school-age population that fails to get into initial learning that is, to acquire reading, writing, and/or calculation correctly, despite normal intelligence and in the absence of gross psycho-affective or socioeducative deficiency. This deficit corresponds to the “specific learning disorder” section of the latest

international classification (DSM-5, 2013). Among these disorders, dyslexia has been the subject of numerous studies in recent years with results clearly demonstrating functional and structural brain abnormalities from both genetic and cultural origins. In short, experiments using brain imaging have shown abnormal activation in several cortical and subcortical brain regions and cerebellum (Démonet et al., 2004) as well as a lack of connectivity between these different areas in children or adults with dyslexia (Finn et al., 2014). This last group of results opens promising new perspectives for understanding the mechanisms underlying dyslexia and related disorders, as well as to guide remediation (van der Mark et al., 2011; Vandermosten et al., 2012). For instance, results of a recent study combining multiple imaging methods (functional magnetic resonance imaging—fMRI—, functional and structural connectivity) revealed that phoneme discrimination impairments, one of the hallmarks of cerebral dysfunction in dyslexia, reflect a failure to access otherwise intact phonemic representations via the subcortical white matter bundles (including the so-called “arcuate fasciculus,” which links Broca’s area to the temporoparietal regions; Boets et al., 2013). The direct implication of this finding is that rehabilitation methods of dyslexia should not only focus on restoring phonological representations, as is the case of most remediations currently used with these children, but also on restoring functional connections between frontal and temporal language areas. More generally, rehabilitation should aim at increasing the integration of information typically processed by different brain areas. As we will argue below, one way to reach this aim is through music training.

Active research in the domain of the neuroscience of music has demonstrated that the brain of professional musicians is an excellent model of brain plasticity (e.g., Münte et al., 2002) both at the subcortical and cortical levels (e.g., Kraus and Chandrasekaran, 2010; Besson et al., 2011). The musician’s brain is ideally suited to study brain changes induced by intensive training and the effects of a targeted and repeated cognitive activity on brain morphology, as suggested for the rehabilitation of dyslexia (Keller and Just, 2009). Interestingly, some white matter subcortical tracts, including the arcuate fasciculus mentioned above and long-known as a crucial element within the left hemisphere language network, are particularly sensitive to learning to play a musical instrument or singing (Halwani et al., 2011), both skills requiring intense and fine coordination between sensory (visual, auditory, and somatosensory) and motor processes. Previous results also demonstrated structural differences in interhemispheric fibers of the anterior region of the corpus callosum connecting motor cortical regions of the right and left hands (e.g., Schlaug et al., 1995). This finding may be related to structural abnormalities of such interhemispheric fibers in children and adults with dyslexia, suggesting that interhemispheric communication failure may be one of the possible mechanisms underlying this disorder (Welcome and Joanisse, 2014).

Using musical training for the remediation of dyslexia and language disorders is based on both theoretical considerations and experimental results. If there are common underlying processes between music and language, especially between music

perception and speech perception, one might assume that improving some of the processes involved in the perception of music can also improve speech perception and reading skills (e.g., Goswami et al., 2002; Patel, 2003, 2012; Kraus and Chandrasekaran, 2010; Besson et al., 2011; Corrigan and Trainor, 2011). In one of the first studies aimed at testing this hypothesis, Overy (2000, 2003) proposed a series of music games gradually increasing in difficulty and focusing on pace and “timing” skills to dyslexic children over a period of 15 weeks. Results showed significant improvements, not in reading skills, but in two related areas: phonological processing and spelling. More recently, Cogo-Moreira et al. (2012, 2013) reported that musical training had positive effects on reading skills and educational achievement in children and adolescents with dyslexia and Weiss et al. (2014) showed that adult musician dyslexics performed better than non-musician normal readers on various pitch interval discrimination tasks, finger rhythmic tapping, and speech in noise perception tasks.

The importance of word metric structure and, specifically, rise-time perception for speech processing has been stressed by Goswami et al. (2002). They proposed that misalignments between neuronal excitability fluctuations in the auditory regions and maximum amplitudes in the speech signal may be related to phonological disabilities in children with dyslexia (Power et al., 2013). In line with this view, Bishop-Liebler et al. (2014) recently reported that adult musician dyslexics were better than non-musician dyslexics on various tests of temporal auditory processing and specifically for processing temporal envelope and “rise time.” In addition, musician dyslexics outperformed their non-musician peers on reading scores and also, to a lesser extent, on phonological awareness. Similarly, Flaugnacco et al. (2014) showed that, among other rhythm production and perception tasks, the level of performance on a metric perception task (i.e., perceiving changes in note duration within recurrent series) specifically predicted both reading speed and accuracy as well as phonological processing in Italian dyslexics. The authors concluded that their results strongly encourage the use of music training in dyslexia rehabilitation, and specifically recommended to “focus on rhythm rather than on pitch accuracy as is often the case in classical music pedagogy.” This recommendation is in line with recent work from the Kraus group (Slater et al., 2013), examining the effect of 1 year musical training based on the perception of pitch, rhythm (tapping in synchrony with a given tempo) and improvisation. The level of performance of 8 year-old children considered “at risk” for learning disability and who received this musical training was significantly higher than matched controls in the synchrony tapping task. Going one step further, Przybylski et al. (2013) examined the influence of rhythm perception on syntactic processing. They presented to language and reading impaired children a rhythmic prime (a succession of notes played either regularly or irregularly), immediately followed by a spoken sentence that was syntactically correct or incorrect (e.g., “Laura *has/have* forgotten her violin”). Results showed a clear superiority for regular over irregular rhythmic primes on the children’s performance in the syntactic task. Based on these results, the authors proposed to use rhythmic stimulation in remediation protocols designed for children with

oral and written language developmental disorders (see also Cason and Schön, 2012; Cason et al., 2015, for similar results with prelingually deaf children).

In a previous work from our group (Chobert et al., 2012), we recorded the Event-related brain Potentials in an original mismatch negativity (MMN) protocol to test for the pre-attentive perception of syllables varying in pitch, duration or voice-onset time. We found that dyslexic children differed from matched controls in the MMN to voice-onset time and to duration but not to pitch variations; that is, to the two time-related among the three variables examined. Directly related to this issue, Bidelman et al. (2014) conducted a series of experiments examining categorical perception, the cornerstone of speech perception, in younger and older adult musicians and non-musicians. Results consistently showed improved categorical perception in musicians than in non-musicians in both younger and older adults. Based on these results and others showing that categorical perception is often impaired in children with dyslexia (Serniclaes et al., 2004), the first aim of the experiments reported here was to further test for categorical perception of voice-onset time (identification and discrimination) in children with dyslexia compared to control normal-readers.

The second aim was to examine perception of word metric structures in children with dyslexia and in normal-readers. We used the materials built by Magne et al. (2007) and comprising trisyllabic words spoken at a normal speech rate or with an unusual lengthening of the penultimate syllable. We hypothesized that children with dyslexia would perform lower than normal readers in the detection of unusual syllabic lengthening.

The third aim was to further test pitch discrimination in dyslexic children since results reported in the literature are quite variable showing both normal or abnormal pitch processing in different studies (e.g., Baldeweg et al., 1999; Santos et al., 2007; Chobert et al., 2012). We included a pitch discrimination task with changes in melodic contour, harmony or both contour and harmony in simple nursery rhymes.

The last and most important aim was to determine whether a specifically-designed Cognitive-Musical Training (CMT) method can improve categorical perception as well as perception of the metric structure of words and possibly pitch discrimination in children with dyslexia. Based on the literature review above, we reasoned that the CMT method should include at least three components: (1) an auditory component targeting the language-music similarity in auditory perception, (2) a motor component, mainly focusing on rhythm production and imitation, and (3) a cross-modal component, making special demands on simultaneous processing of information from different modalities including auditory, visual, sensory, and motor modalities as well as their combinations. To this end and in order to stimulate auditory attention and working memory abilities, the CMT program comprises a battery of musical tasks and exercises based on these components and on the active listening to various sorts of musical stimuli. Multimodal training combining different modalities, as is typically the case in real life, was achieved through tasks that simultaneously involve visual, auditory, and sensory-motor processing. Simplified visual and

gestural supports were provided that were adapted to each child's level of performance. For instance, a very simple musical notation system was purposefully devised, made of only 3 or 5 strokes to represent pitch and duration of sounds. The rhythmic aspect of each task was emphasized (for details regarding the content of exercises and tasks, see the Methods Section below and Habib and Commeiras, 2014).

Two experiments were conducted with two different populations of dyslexic children, using similar tasks and materials and only differing in the duration of the musical training period. Thus, we compared the effects of the CMT program when training sessions were clustered on 3 consecutive days and when they were distributed over a period of 6 weeks.

STUDY 1: INTENSIVE COGNITIVE-MUSICAL TRAINING (CMT) CLUSTERED ON 3 CONSECUTIVE DAYS

Methods

Participants

A group of 12 children from 8.2 to 11.7 years (mean 10 years 7 months, *s.d.* = 17 months) participated in the study. They all received a common diagnosis of severe dyslexia leading to their admission in specialized classes with multi-disciplinary support for dyslexic children. Thus, all children were already involved in intensive conventional rehabilitation methods, but during this 3-day CMT period, they did not receive speech therapy. The clinical characteristics of the children are reported in **Tables 1, 2**. A reading-age matched normal-reading group of 22 children (30 months younger on average), served as a control population for normative data. As seen from **Tables 1, 2**, the diagnosis of severe dyslexia rests on the presence of significant delay in terms of reading age as well as reading isolated words. General intelligence was largely preserved, as shown by scores on the similarities and matrices subtests of the WISC-IV scale. Spelling and auditory-verbal short-term and working memory were more variably altered.

Training Procedure

Description of the CMT Method

The CMT method was designed by speech therapists based on widely recognized principles of effective intervention (i.e., goal-directed, systematic, and coherent progression along a hierarchical structure; Shaywitz, 2005). Several exercises were built that covered various dimensions and components of music: pitch, duration, tempo, pulsation, and rhythm and that aimed at developing both the perception and the production sides. Exercises also comprised both sensory (visual and auditory) and motor components, engaging the child into transcoding processes from one modality to another (e.g., tapping in synchrony with a heard sequence, tapping the written notation of a rhythm; learning to play a small melody and to correct errors in other children's performance; etc...). The use of a piano keyboard was systematically added to provide children with the visuo-spatial organization of the white and black keys, as a reinforcement for the sequential nature of the musical

TABLE 1 | Level of performance of dyslexics and control normal-readers (with indication of two standard deviations below norm) in several standard psychometric tests.

		Dyslexics		Controls	
		Mean	±SD	Mean	−2 SD
Reading age		90.0	±19.0	128.5	93.9
Oral and written language battery	Phonetic fluency	15.0	±2.8	15.0	8.0
	Semantic fluency	27.3	±9.0	23.5	16.0
	Hard words repetition	23.2*	±5.0	29.5*	26.0
	Reading strategy				
	Pseudowords	15.3*	±2.4	20.0*	16.0
	Regular	9.1	±1.2	10.0	9.4
	Irregular	6.8*	±1.9	10.0*	8.0
	Spelling				
	Phonological errors	11.3*	±3.0	15.0*	12.0
	Grammatical errors	10.0	±7.0	9.5	4.5
	Use rules errors	14.0	±5.9	18.5	9.5
WISC-IV	Similarities	10.4	±2.4	9.7	4.9
	Matrices	10.1	±3.0	10.3	4.3
	Digit span				
	Direct	7.8	±2.1	9.9	3.5
	Reverse	7.9	±3.4	9.9	4.7
TOTAL		7.9	±2.6	9.9	4.5

Significant control/patient differences are in bold (T-test, * $p < 0.05$). Reading ages are in months.

TABLE 2 | Dyslexia severity (age in month).

Dyslexics	Chronological age	Reading age	Difference
1	135	111	−24
2	133	90	−43
3	146	111	−35
4	146	88	−58
5	99	55	−44
6	97	77	−20
7	125	84	−41
8	112	58	−54
9	141	101	−40
10	124	106	−18
11	123	96	−27
12	145	114	−31
Mean	127	90	−36
s.d.	17	19	12

scale. Also and as often as possible, exercises required body movements to be performed in line with the musical excerpts. Finally, the connection between music and language was used through exercises implicating both speech and music (e.g., nursery rhymes, tracing the prosody of a sentence on a sheet of paper...). During the CMT sessions, no speech therapy was used

and no conventional exercises (e.g., related to phonology reading or writing) were performed. However, most children were also involved in their usual weekly treatment, more or less half an hour, one or two times per week with a speech therapist.

The CMT program started on the first day of winter vacation in an outbuilding of the University Hospital, after the children and their legal representatives had agreed to participate in this experiment that was approved by the *ad-hoc* local ethics committee. They were fully informed of the aim and content of the research program. The training workshop lasted for 3 whole days, 6 h per day, for a total of 18 h. Children were divided into three groups of four and they all participated in three training sessions including (1) specific musical exercises given by a speech therapist, (2) music education with piano instruction with a piano professor, and (3) percussion and rhythmic bodily exercises with a psychomotor therapist. Each session lasted 45 min, with a 15 min break before moving onto the next training session. Each of the 3 days included the same sequence of sessions, only varying in level of difficulty. At the end of each day, all children met in a dance hall, where they practiced folk dancing with a specialized teacher. Children were informed that they will perform in front of their parents and teachers at the end of the third and final day to give a more recreational and challenging aspect to the whole training and to increase their motivation.

Assessment Tests

As described below, three tasks tapping into different aspects of auditory and speech perception, were used: categorical perception (identification and discrimination tests), syllabic duration and pitch variations. The level of performance in each task was measured in children with dyslexia both before and after training. For comparison purposes, the level of performance in each task was also measured in control normal-readers who did not follow the CMT program as they were not impaired in the perceptual, cognitive, and motor abilities that the CMT aimed at improving.

Categorical Perception of the Phoneme “b” in the Syllable [Ba]

Identification Task

A 9-step continuum between two phonemes (i.e., “b” in syllable “Ba” from B1 to B4 and “p” as in syllable “Pa” from B5 to B9) was used with voicing delays varying between −52 and +20 ms. The identification rate of Ba (that of Pa being represented by the reverse curve) was computed.

Discrimination Task

Eight pairs were formed (from Ba1–Ba2 to Pa8–Pa9) using the nine syllables of the continuum. The rate of correct discrimination (same-different) was computed.

Syllabic Duration Task

Children listened to 42 tri-syllabic words (e.g., “canapé”) and they had to decide whether the word was spoken normally or with an incongruous lengthening of the penultimate syllable (e.g., “canaapé”). The percentage of correct responses was measured. **Pitch discrimination task:** children listened to five nursery

rhymes played on the piano (e.g., “Sur le pont d’Avignon”; each around 20 s duration) that were recorded in four different versions: exact version, pitch change within the melodic contour, pitch change out of melodic contour, pitch change out of melodic contour and out of harmony, for a total of 20 trials. Children were asked to decide whether each fragment was the normal version or not.

Data Analysis

First, we compared dyslexics and normal-readers in the different tasks (identification and discrimination tasks for categorical perception, metric, and pitch discrimination tasks) using Analyses of Variance (ANOVAs) including Group (Dyslexics vs. Controls) as a between-subject factor and Position on the continuum (nine Positions; identification task), Pairs (eight different pairs; discrimination task), Syllabic duration (normal vs. lengthened; metric task), or Pitch (normal pitch, pitch change preserving melodic contour, pitch change out of melodic contour, and out of harmony) as within-subject factors in separate ANOVAs. To test for the effects of the CMT program in dyslexic children, we also computed ANOVAs including Session (before vs. after CMT) as well as the within-subjects factors described above for each task. Simple effects were analyzed using *post-hoc* Fischer’s PLSD tests. All analyses were computed using the *Statistica* program.

Results

Categorical Perception

As shown in **Figure 1A**, the percentage of syllables identified as “Ba” differed between children with dyslexia before CMT and normal-readers. However, while, the main effect of Group was not significant [$F_{(1, 29)} = 2.16, p = 0.15$], the main effect of Position [$F_{(8, 29)} = 193.61, p < 0.001$] and the Group \times Position interaction [$F_{(8, 232)} = 6.97, p < 0.001$] were significant. *Post-hoc* analyses showed that the between-group differences were significant for position B2 ($p < 0.007$), with a higher percentage of “Ba” identification for dyslexics than controls as well as for B5 ($p < 0.03$), B8 ($p < 0.02$), and B9 ($p < 0.002$), with a lower percentage of “Ba” identification for dyslexics than for controls.

For the discrimination task (see **Figure 1B**), both inter- and intra-categorical pairs were presented (the latter being harder to discriminate). Neither the between-group difference ($F < 1$) nor the Group \times Pairs interaction ($F < 1$) were significant. Only the main effect of Pairs was significant [$F_{(7, 33)} = 5.56, p < 0.001$] with the highest percentage of correct discrimination for the B4–B5 pair (Pair 4 on **Figure 1B**) for all children.

Testing for the effect of the CMT program, results of separate ANOVAs for dyslexics in the identification task showed significant improvements after 3 days of CMT with no main effect of Session ($F < 1$) but a significant main effect of Position [$F_{(1, 11)} = 68.94, p < 0.001$] and a significant Session \times Condition interaction [$F_{(8, 88)} = 2.41, p < 0.021$]. *Post-hoc* analyses showed that both within-category and inter-category perceptions were modified after training (B2: $p < 0.03$; B6: $p < 0.03$; and B9: $p < 0.04$).

Results for dyslexics in the discrimination task revealed that the main effect of Session was marginally significant [$F_{(1, 11)} =$

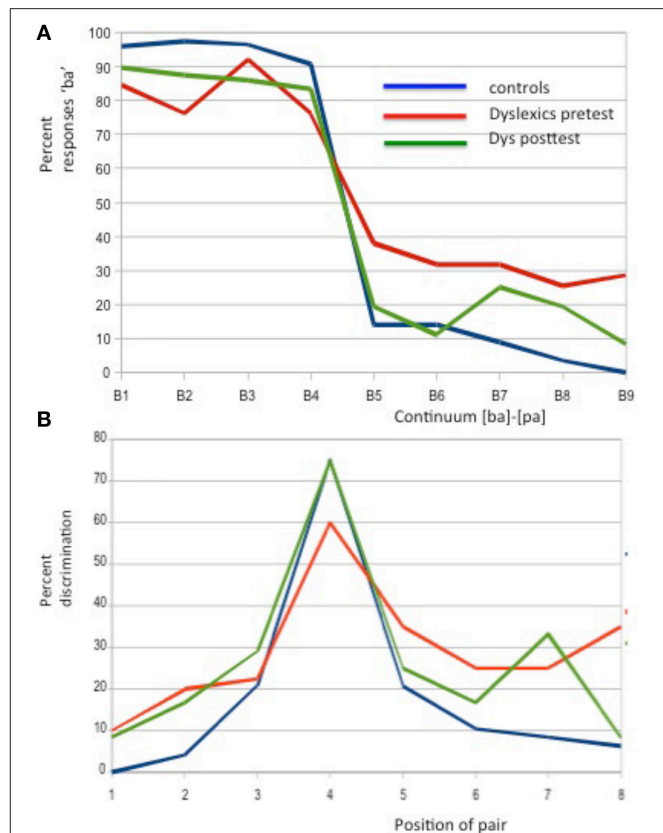
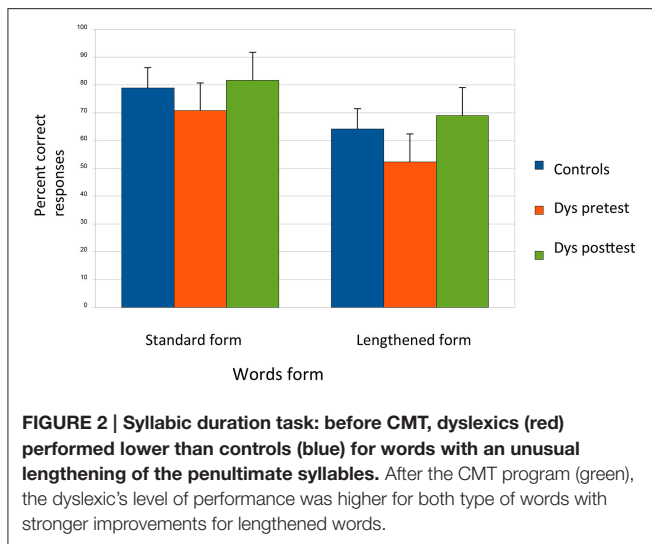


FIGURE 1 | Categorical perception using a nine steps continuum between the syllables [ba] and [pa]. In the identification test (**A**) dyslexic children before CMT (red) showed less steep intercategorical boundary than normal readers (blue) but a “normalization,” specifically for B5 and B6 after CMT (green). In the discrimination task (**B**), dyslexics before CMT (red) seemed to differ from normal readers (blue) for items at or close to the inter-categorical border (median peak in the figure) but these differences vanished after CMT (green).

3.61, $p < 0.08$] but the main effect of Pairs as well as the Session by Pairs interaction were significant [$F_{(1, 11)} = 16.32, p < 0.001$ and $F_{(7, 77)} = 3.28, p < 0.004$]. The improvement in phonetic discrimination after CMT was largest for the pairs 4 and 5. *Post-hoc* analyses confirmed both intra (pairs B1–B2; $p < 0.02$) and inter (B5–B6, $p < 0.007$) category perception improvement.

For the Syllabic Duration Task (Figure 2)

The level of performance of dyslexics before training was significantly lower than for normal readers [main effect of Group: $F_{(1, 33)} = 4.97, p < 0.03$]. Moreover, all children performed lower for words with lengthening of the penultimate syllables than for normally spoken words [main effect of Condition: $F_{(1, 33)} = 27.21, p < 0.001$] and the Group \times Condition interaction was also significant [$F_{(1, 33)} = 4.2, p < 0.05$]. *Post-hoc* comparisons showed that dyslexics (52%) performed lower than controls (64%) for words with lengthening of the penultimate syllables with no differences for normally spoken words (dyslexics: 73% and controls: 78%).



Importantly, results of separate ANOVA for dyslexics, showed that the main effects of Session and Syllabic duration were significant [$F_{(1, 11)} = 16.62, p < 0.001$ and $F_{(1, 11)} = 8.96, p < 0.01$ respectively]. The Condition \times Session interaction was only marginally significant [$F_{(1, 11)} = 3.15, p = 0.10$]: the level of performance of dyslexics was higher after CMT than before for both normally spoken words (after: 81% and before: 73%) and for lengthened words (after: 69% and before: 52%). Nevertheless, results of *post-hoc* tests showed that the improvement was larger for lengthened words ($p < 0.002$) than for normally spoken words ($p < 0.02$).

For the Pitch Discrimination Task (Nursery Rhymes)

Results revealed that dyslexics did not differ from controls before CMT [main effect of Group: $F_{(1, 32)} = 1.36, p > 0.25$]. The main effect of Condition was significant [$F_{(3, 96)} = 89.35, p < 0.001$] but the Group \times Condition interaction was only marginally significant [$F_{(3, 96)} = 2.34, p = 0.07$]. The differences between dyslexics and controls were larger for the exact version condition ($p < 0.05$) than for the other three conditions (all $p > 0.50$).

Results of separate ANOVA comparing dyslexic children before and after the CMT program showed no main effect of Session ($F < 1$) and no Session \times Condition interaction ($F < 1$). The overall percentage of correct responses of dyslexic children was not higher after (56, 30, 26, 30%) than before CMT (54, 30, 30, 29%). By contrast, the main effect of Condition was significant [$F_{(3, 33)} = 18.06, p < 0.001$]. Results of *post-hoc* tests showed that the level of performance was highest for the “normal” condition ($p < 0.002$).

Discussion

Results of this first experiment that aimed at testing the effects of an intensive use of the CMT method over 3 consecutive days in children with dyslexia, revealed two findings of main interest. First, compared to normal-readers, dyslexics were impaired in the identification test of categorical perception but their level of performance reached the level of control children after 3 days (18 h) of the CMT program. That dyslexics and controls were

significantly different in the identification task before training suggests an excessive intra-categorical and less clear inter-categorical perception. Importantly, intensive music training positive influenced categorical perception by facilitating syllabic identification based on differences in VOT between the “b” and “p” phonemes. Likewise, there was a significant improvement in the discrimination test of categorical perception wherein intra-categorical pairs were more often perceived as different by dyslexics than by normal-readers, possibly reflecting some type of allophonic perception (Serniclaes et al., 2004). Overall, these results are in line with improved perception of VOT with music training in normal readers (e.g., Chobert et al., 2014) and with vowel identification in young and older musician compared to non-musician adults (Bidelman et al., 2014).

The present results also showed that while the level of performance of children with dyslexia before CMT was lower than normal readers in the syllabic lengthening task, it was significantly improved after 3 days of CMT. These findings are in line with those of a previous study using the same stimuli and showing that musicians were more sensitive than nonmusicians to the abnormal lengthening of the penultimate syllable of trisyllabic words (Marie et al., 2011). More generally, these results support the view that deficits in children with dyslexia are linked to temporal processing of speech insofar as time-dependent variables such as VOT and duration are the most altered (Goswami et al., 2002, 2013). In this respect, the improvement found after music training in the children tested here possibly resulted from the CMT focusing on the manipulation of the temporal characteristics of sounds: rhythm and tempo for non-speech sounds and duration or voicing for speech sounds. Finally, the present results showed no deficits in pitch discrimination in dyslexics (e.g., Chobert et al., 2012), unlike previous evidence of the contrary (e.g., Baldeweg et al., 1999; Santos et al., 2007), and no improvement after CMT. However, this is to be expected since the CMT focussed on the rhythmic and temporal aspects of music training.

In sum, these results were encouraging in showing a positive effect of the CMT program on auditory-verbal variables that were not specifically trained after 3 days (18 h) of intensive intervention. While the several caveats present in this experiment (e.g., lack of an appropriate control group, multi-dimensionality of the MT program...) will be considered in the general discussion, we first present the second experiment aimed at testing the effects of this type of intervention conducted over a longer period. To this aim, the CMT program was used with a different group of children with dyslexia trained over 6 weeks. We used the same tasks as in Experiment 1 together with several standardized psychometric tests of various cognitive functions.

STUDY 2: COGNITIVE-MUSICAL TRAINING OVER 6 WEEKS: ANALYSIS OF THE EFFECTS ON A BATTERY OF COGNITIVE AND SPEECH TESTS

Many questions remained unanswered after Experiment 1. First, it was of importance to determine whether the effects observed

after three CMT training days could be replicated in conditions more compatible with regular primary school schedule, so that it can be applied in current practice by speech therapists or other specialists. Second, it was of interest to test whether the observed effects generalized to variables directly involved in the nature of learning difficulties such as phonology, reading, or spelling. Third, one could question the sustainability of the observed effect since it would lose interest if it only proved ephemeral.

To answer these questions, we used a CMT similar in content and total duration but spread over 6 weeks and in a different context, that of a classroom of 12 dyslexic children, all of them with a main diagnosis of severe dyslexia. We took advantage of the existence, in the Marseille area, of schools providing special classrooms for dyslexic children (i.e., “CLIS-DYS”: sections for school inclusion of dyslexics). In contrast to the previous study, the experimental design involved three 6-week periods, including two untrained periods, one before (between T1 and T2) and one after (between T3 and T4) the CMT period (between T2 and T3). Measurements were taken four times, before and after each period (i.e., T1, T2, T3, and T4).

We hypothesized that the children’s level of performance in auditory, phonological, and reading tasks, but not in writing and visual tasks, would specifically improve during the CMT period that is between T2 and T3. Any improvement between T1 and T2 (i.e., before the start of CMT) would suggest the influence of other, confounding factors. Moreover, the lack of significance for T4 vs. T3 comparisons would be compatible with the persistence of the beneficial effects beyond the end of CMT.

Methods

Participants

A total of 12 children were grouped according to the intensity of their problems and not on age. Indeed, their age difference prompted us to work on homogeneous groups of four children based upon school criteria provided by the teaching team:

- one group that just stepped into reading: 4 boys aged 7, 9, 10, and 11 years.
- one mid-level group who did not yet reach automation in reading: two girls 9 and 10 year-old, and two 10 year-old boys.
- one group who had reached automation in reading: two girls aged 11 and two boys aged 11 and 12.

Training Protocol

All children were participating in workshops that took place during school time as detailed below, 3 h per week for 6 weeks. Within each of the 6 weeks, four interventions took place: two workshops of 1 h of CMT in full class (12 children) provided by a speech therapist and two musical workshops in smaller groups (4 children) for half an hour, including piano and percussion practice. Although they differed in mean age, the four groups basically received the same type of intervention, with similar content but with the easiest exercises for the youngest children. The content of the training was similar to Experiment 1, except for the dancing activity which was not proposed in Experiment 2.

Assessment Battery

Efficiency of the CMT method was assessed using a large battery of language and reading tests as well as other psychometric tests focusing on rhythm, auditory attention, visuo-spatial attention, sequential visual processing, phonological awareness, speed and quality of reading, audio-vocal loop in working memory. Moreover, the identification and discrimination tasks as well as the syllabic duration task used in Experiment 1 were also presented here. The tests were performed four times: at T1, 6 weeks before the start of the workshops; at T2 and T3, just before and just after the end of the workshops, respectively, and at T4, 6 weeks after the end of the workshops. Due to technical problems, the categorical perception and syllabic duration tasks were only performed twice, at T2 and T3 that is, just before and after CMT training.

Language and Cognitive Tasks

Three tasks were selected from the NEPSY II Battery (“A Developmental NEuroPSYchological Assessment”; Korkman et al., 2012).

- **Auditory Attention and Response Set** Children are listening to a pre-recorded tape. Part A: when they hear the word “red” they put a red square into a box and when they hear other words, they do nothing. Part B: when they hear the word “red” they put a yellow square in the box and when they hear the word “yellow,” they put a red square in the box. When they hear the word “blue,” they put a blue square in the box. These tasks allow testing for selective and sustained attention as well as for executive function, specifically inhibition, and shifting.
- **Visuo-Spatial Attention** Children are required to cross-over as quickly as possible a specific symbol among hundreds of other symbols presented on a sheet of paper. Part B: Children are required to cross-over as quickly as possible two specific symbols among hundreds of other symbols. The level of performance is computed as the number of false alarms subtracted from the number of correct responses. Time is limited to 180 s per sheet. These tasks allow testing for perceptual attention and visuo-motor abilities.
- **Repetition of Non-sense Words** Children are asked to repeat nonsense words presented from an audiotape. These tasks allow testing for phonological encoding and decoding skills.

Four tasks were chosen from the BALE battery (“Batterie Analytique du Langage Ecrit”; <http://www.cognisciences.com>) that allow testing for reading, spelling, and meta-phonological skills in French.

- **Digit Repetition Task** Children are asked to repeat sequences of digits that increase in size (forward span) or to repeat the sequences of digits in reverse order (backward span) until they make two consecutive errors. These tasks test for short-term and working memory.
- **Phonemic Fusion** Children are asked to isolate the first phonemes of two consecutively presented words and to merge them together to create a syllable [e.g., the answer for “bel animal” (“beautiful animal”) is [ba]]. Three examples are given before starting. Ten items are presented, the number of correct responses is computed and the time taken to perform the task is measured.

- **Visual Identification of Letters (Sequential Analysis)** Twenty series of 3–5 letters are presented in pairs and columns and children have to decide whether both members of the pair are similar or not.
- **Contour Discrimination** Using four different color pens, children have to highlight the contour of four intermixed stars. This task tests for perceptual discrimination.
Finally, three standardized task were used to test for reading abilities, rhythm reproduction and writing.
- **Reading Task** (“Lecture en une Minute,” LUM; 1 min Reading Task; LMC-R Battery, Khomsi, 1999) Children are asked to read as many words as possible that are presented in a column. The number of words read in 1 min and the number of errors are recorded and the difference between the two measures gives the reading score in 1 min (LUM). This test evaluates the degree of automation in reading, a key element of reading efficiency.
- **Rhythm Reproduction Task (Stambak, 1951)** Children are asked to reproduce a set of 21 rhythmic patterns of increasing complexity that are performed by the examiner following indications on a sheet of paper. Scores are computed by counting the number of errors in the reproduction of the rhythmic patterns.
- **BHK Test (Concise Evaluation Scale for Children’s Handwriting, French Version, Charles et al., 2003)** Children are asked to copy a standard text that is presented on a card for 5 min. This task tests for the quality and fluidity of handwriting. Results are scored according to 13 different criteria (such as size, regularity, variations in size or obliquity of letters, etc. . .) and a separate criterion of writing speed.

Data Analysis

To allow for comparisons with Experiment 1, repeated measures ANOVAs were conducted for the categorical perception tasks that included Position or Pair and Session (T2 vs. T3) as within-subject factors. Fischer’s PLSD were used for *post-hoc* comparisons. For the syllabic duration task, the ANOVA included Condition (normal vs. lengthened syllables) and Session (T2 vs. T3) as within-subject factors. Student *t*-tests were used for pre vs. post-test comparisons in the other tests. When possible, scores were transformed into standard deviation units from the norm (as provided by authors of the tests). Means, standard deviations and significance level for each individual measurement are reported in **Tables 3–5**. When relevant, effect sizes are also reported as Cohen’s *d* (Cohen, 1988; Soper, 2015). Finally, in order to correct for multiple comparisons and considering that <10 independent planned comparisons were computed, the significance level was set at $p < 0.01$ rather than $p < 0.05$.

Results

A- Categorical Perception and Syllabic Duration Tasks

Categorical perception

For the identification task (**Figure 3A**), results revealed that both the main effect of Position and the Session \times Position interaction were significant [$F_{(1, 11)} = 35.58, p < 0.001$ and $F_{(8, 88)} = 2.50,$

$p < 0.01$, respectively] but the main effect of Session was not significant ($F < 1$). *Post-hoc* comparisons showed that the improvement from T2 to T3 was significant for positions B5 and B6 (i.e., around the phonemic [ba]-[pa] boundary with $p < 0.03$ and $p < 0.004$, respectively).

For the discrimination task (**Figure 3B**), the main effect of Pair was significant [$F_{(1, 11)} = 4.46, p < 0.001$] and the main effect of Session was marginally significant [$F_{(1, 11)} = 2.88, p < 0.11$]. The Session \times Position interaction was not significant [$F_{(7, 77)} = 1.39, p = 0.21$]. However, *post-hoc* analyses revealed that the improvement from T2 to T3 was significant for the B4/B5 pair ($p < 0.02$) and for the B5/B7 pair ($p < 0.004$).

Syllabic duration task

The main effect of Condition was not significant [$F_{(1, 11)} = 1.50, p = 0.24$] so that lengthened words were not processed differently than normally spoken words. The main effect of Session was only marginally significant [$F_{(1, 11)} = 2.42; p < 0.14$]. The Session \times Condition interaction was not significant ($F < 1$) and *post-hoc* analyses also revealed marginal improvements between T2 and T3 for both normally spoken words ($p = 0.10$) and words with syllabic lengthening ($p = 0.08$).

B- Attentional Processing of Speech and Non Speech Stimuli

Auditory attention

Results for the two auditory attention subtests from the NEPSY battery showed that total performance ($t = -5.72, p < 0.001$) and performance in both subtests (A: $t = -6.35, p < 0.001$) and (B: $t = 4.25, p < 0.01$) were improved from T2 to T3 with no decrease from T3 to T4 (all $p > 0.15$) and with no difference between T1 and T2 (all $p > 0.70$; see **Table 3**).

Visuo-spatial attention

No significant improvement was found from T2 to T3, but performance improved from T1 to T2 when children received no treatment (T1-T2: $t = 3.77, p < 0.003$; see **Table 3**).

Digit repetition task from the battery BALE

No significant improvement was found whatever the period considered (all $p > 0.30$, see **Table 3**).

Reproduction of motor rhythmic sequences

No significant differences were found whatever the period considered (all $p > 0.10$, see **Table 3**).

C- Phonological and Reading Tasks

Pseudo-word repetition

Results showed improvements from T2 to T3 ($t = -2.56, p < 0.026$), no decrease from T3 to T4 ($p > 0.15$) and no difference between T1 and T2 ($p > 0.80$, see **Table 4** and **Figure 4**).

Reading in 1 min (Khomsi test)

Results showed improvements from T2 to T3 ($t = -5.59, p < 0.001$) with no significant decrease from T3 to T4 ($p > 0.80$) and no significant difference between T1 and T2 ($p > 0.20$, see **Table 4**).

TABLE 3 | Attentional processing of speech and non speech stimuli.

Task	Type of measure	T1 mean	T2 mean	T3 mean	T4 mean	T1/T2		T2/T3			T3/T4		
		(s.d.)	(s.d.)	(s.d.)	(s.d.)	T-test	P-value	Effect size	T-test	P-value	Effect size	T-test	P-value
Auditory attention	Correct resp. (A)	32.83 (19.30)	31.33 (5.15)	45.16 (6.71)	45.25 (2.85)	0.29	0.77		6.35	0.001***	2.31	0.04	0.96
	Correct resp. (B)	29.25 (12.31)	30.16 (6.64)	38.80 (8.58)	38.50 (8.63)	0.39	0.70		4.25	0.01**	1.12	0.14	0.89
	Total (s.d. from norm)	-1.0 (0.83)	-1.00 (0.31)	-0.36 (0.54)	-0.25 (0.62)	0.01	0.99		5.72	0.001***	3.09	1.48	0.17
Visuo-spatial attention	Correct resp. (A)	19.66 (1.15)	19.91 (0.28)	2.00 (0.00)	19.91 (0.28)	0.71	0.49		1.00	0.33		1.01	0.33
	Correct resp. (B)	14.00 (3.83)	16.90 (3.20)	17.00 (2.76)	16.60 (3.11)	-3.77	0.003**	0.82	0.16	0.87		0.84	0.42
	Total (s.d. from norm)	-0.86 (1.0)	-0.50 (0.77)	-0.69 (1.12)	-0.72 (1.03)	1.77	0.10		0.50	0.62		0.11	0.91
Working memory: Digit span (s.d. from norm)	Forward (s.d. from norm)	1.51 (1.10)	1.22 (1.05)	1.13 (0.98)	0.87 (1.36)	-1.08	0.30		0.44	0.67		0.68	0.51
	Backward (s.d. from norm)	-1.07 (0.52)	-0.99 (0.47)	-0.83 (0.63)	-0.59 (0.88)	0.40	0.68		0.75	0.47		1.04	0.32
Rhythm reproduction	Errors	1.16 (3.07)	9.58 (2.81)	8.25 (2.17)	9.33 (2.08)	0.61	0.55		1.34	0.21		-1.82	0.09

Children' levels of performance were measured four times (T1, 6 weeks before CMT started; T2, just before CMT; T3, just after CMT; T4, 6 weeks after CMT ended). Mean and standard deviation or s.d. from norms (when indicated) for each task are reported. Student T-tests and p-values were computed; significant improvements are in bold (** $p < 0.001$; ** $p < 0.01$). Effect sizes are also reported when relevant (Cohen's d).

TABLE 4 | Phonological and reading tasks.

Task	Type of measure	T1 mean	T2 mean	T3 mean	T4 mean	T1/T2		T2/T3			T3/T4	
		(s.d.)	(s.d.)	(s.d.)	(s.d.)	T-test	P-value	T-test	P-value	Effect size	T-test	P-value
Pseudo-word repetition	Pseudo-word span	21.08 (8.83)	21.5 (6.20)	24.75 (6.16)	26.50 (6.15)	0.21	0.84	2.57	0.03*	0.52	1.48	0.17
Reading in 1 min (LUM)	Nb items read (s.d. from norm)	-2.24 (1.32)	-2.12 (1.49)	-1.66 (1.59)	-1.64 (1.64)	1.34	0.20	5.59	0.001***	0.29	0.20	0.85
Phoneme fusion (s.d. from norm)	Phonemic fusion score	-0.86 (0.96)	-0.58 (0.86)	0.04 (0.73)	-0.05 (0.80)	1.04	0.32	2.90	0.01**	0.78	0.70	0.50
	Time phoneme fusion	-0.31 (1.19)	-0.29 (1.15)	-0.02 (1.13)	0.06 (0.86)	0.32	0.75	1.94	0.07		0.46	0.65

Children' levels of performance were measured at T1 (6 weeks before CMT started) at T2 (just before CMT), at T3 (just after CMT), and at T4 (6 weeks after CMT ended). Mean and standard deviation or s.d. from norm (whenever indicated) are reported for each task. Student T-tests and p-values were computed and significant improvements are in bold (** $p < 0.001$; * $p < 0.01$; * $p < 0.05$). Effect sizes are also reported when relevant (Cohen's d).

Phoneme fusion (BALE)

Results showed an improvement from T2 to T3 for accuracy ($t = -2.90$, $p < 0.01$), with only a tendency for speed ($t = 1.94$, $p = 0.07$). No decrease was found from T3 to T4 whether for accuracy or for speed (all $p > 0.40$) and no difference was found between T1 and T2 (all $p > 0.30$; see **Table 4** and **Figure 4**).

D- Visual and Writing Tasks

Comparison of letter strings (BALE)

The improvement from T2 to T3 was significant for speed ($t = 3.41$, $p < 0.006$) but only marginally significant for accuracy ($t = -1.78$; $p = 0.10$). No significant decrease was found from T3-T4 ($p > 0.40$ in both cases) and no significant

difference between T1 and T2 ($p > 0.20$ in both cases, see **Table 5**).

Contour discrimination (BALE)

The improvement from T1 to T2 was significant ($t = 2.55$, $p < 0.03$) with no change from T2 to T3 or from T3 to T4 (see **Table 5** and **Figure 5**).

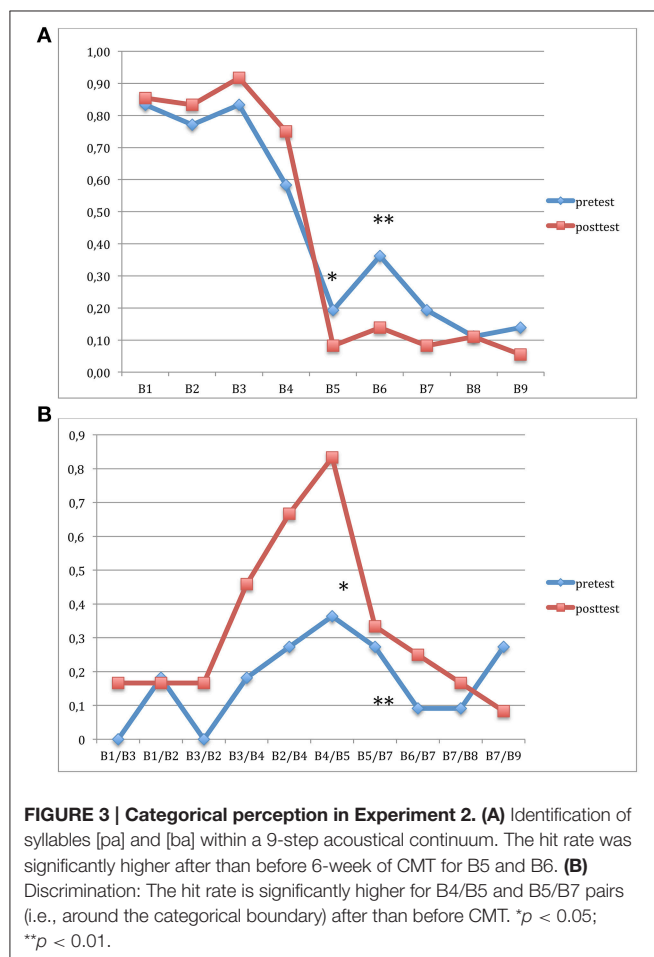
BHK (writing)

The improvement from T3 to T4 was marginally significant ($t = -2.17$, $p < 0.05$) with no change from T2 to T3 or from T1 to T2 ($p > 0.15$, see **Table 5**).

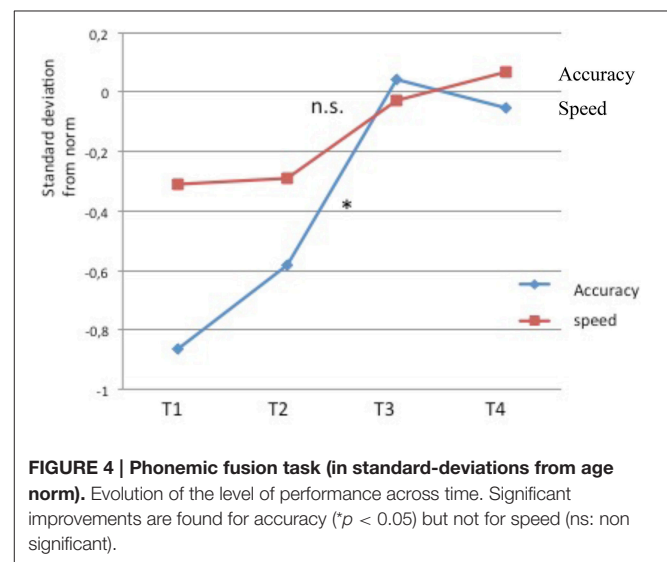
TABLE 5 | Visual and writing abilities.

Task	Type of measure (s.d.) from norm)	T1 mean (s.d.)	T2 mean (s.d.)	T3 mean (s.d.)	T4 mean (s.d.)	T1/T2		T2/T3		T3/T4	
						T-test	P-value	Effect size	T-test	P-value	Effect size
Letter-sequence comparison	Score	-1.27 (1.74)	-2.21 (2.98)	-0.55 (0.90)	-0.46 (1.07)	1.01	0.33		1.78	0.10	
	Time	-1.54 (1.24)	-1.36 (1.22)	-0.40 (0.50)	-0.28 (0.72)	1.31	0.21		3.41	0.006**	1.02
Contour discrimination	Nb correct contours	-4.97 (6.25)	-1.44 (3.31)	-1.44 (3.84)	-1.44 (3.84)	2.55	0.03*	0.70	n.a.		n.a.
Writing test BHK	Score quality	-1.84 (2.37)	-1.70 (2.36)	-1.19 (2.15)	-1.36 (2.51)	0.43	0.67		1.08	0.30	
	Speed	-1.65 (0.61)	-1.55 (0.60)	-1.67 (0.67)	-1.28 (0.80)	0.98	0.18		1.46	0.17	2.17

Children' levels of performance were measured at T1 (6 weeks before CMT started) at T2 (just before CMT), at T3 (just after CMT), and at T4 (6 weeks after CMT ended). Standard deviation from norm for each task are reported. Student T-tests and p-values were computed and significant improvements are in bold (** $p < 0.01$; * $p < 0.05$). Effect sizes are also reported when relevant (Cohen's d). n.a.: not available.



Finally, correlational analyses between the different tasks did not reveal any significant results when the significance level is set at $p < 0.01$.



Discussion

The first aim of Experiment 2 was to determine whether results similar to Experiment 1 would be found when CMT was spread over time. The second aim was to assess whether or not this effect also extended to standardized psychometric tests known to be sensitive to learning difficulties encountered by children with dyslexia. Overall, these two objectives were reached. As in Experiment 1, results revealed improved categorical perception of syllables after the period of CMT (significant Session \times Condition interaction in both experiments). While improvements were found both for the quality of intra-categorical perception and for inter-categorical boundary in Experiment 1 (i.e., at B2, B6 and B9), the improvement was mainly found for inter-categorical boundary in Experiment 2 (i.e., at B5 and B6). Nevertheless, in both experiments and in line with previous results in younger and older adults (Bidelman et al., 2014), music training seemed to positively influence categorical

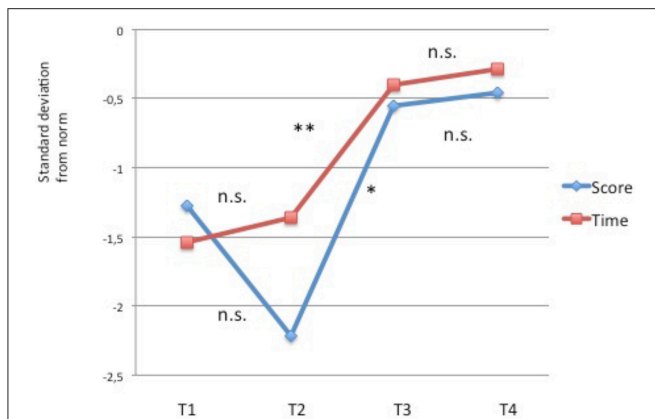


FIGURE 5 | Comparison of letter strings (in standard deviations from age-norm). Evolution of the level of performance across time. Significant improvements from T2 to T3 for speed (Time: ** $p < 0.01$; * $p < 0.05$) but marginally significant for accuracy (score: * $p < 0.05$).

perception in children with dyslexia. Turning to the perception of syllabic duration, the Session by Condition was marginally significant in both experiments. While the effect of CMT was larger for lengthened than normally spoken words in Experiment 1, the perception of both types of words was marginally improved after the CMT in Experiment 2. Thus, overall results were similar in both Experiments, showing that the CMT program positively influenced categorical perception and the temporal aspects of speech processing. Importantly, these effects were found in Experiment 2 when the CMT program was spread over time and, consequently, more compatible with standard speech therapy practice.

Results of standardized psychometric tests showed that several aspects of the children's behavior that were directly targeted by the CMT program specifically improved during the period of music training. This was clearly the case for auditory attention, pseudo-word repetition, reading words in 1 min, phonological awareness (phonemes fusion), and comparison of letter strings. Specifically, for auditory attention, results showed almost 15% gain in selective attention, 10% in divided attention, and 20% in total score from T2 to T3. Importantly, these improvements persisted in the following period without further training (from T3 to T4: no significant decrease in level of performance). Turning to the reading tests that were probably the most interesting due to their strong relationship to academic performance, the improvement after CMT was almost one standard deviation from the norm, moving from a score lower to -2 s.d. to nearly -1 s.d. from controls scores. Similar to results for auditory attention, pseudo-word repetition and phoneme fusion (scores), these reading improvements persisted unchanged for 6 weeks after the end of the CMT period, thereby pointing to the durability of the CMT program.

Equally important, the effect of the CMT program was not significant between T2 and T3 on the control variables for which we did not *a priori* predict an effect of this type of treatment, such as visuo-spatial attention, contour discrimination, and

writing efficiency. However, some effects, such as the significant improvement between the untrained period from T1 to T2 for visual attention and contour discrimination and from T3 to T4 for writing efficiency, were unexpected and may result from mere repetition effects. They need to be replicated and examined in further experiments. More surprisingly in view of several results in the literature showing strong links between rhythmic and linguistic abilities (Overy, 2000, 2003; Przybylski et al., 2013; Slater et al., 2013; Bishop-Liebler et al., 2014; Flaughnacco et al., 2014; Weiss et al., 2014) and in view of the strong focus of the CMT program on the temporal association between sensory input and motor activities, we found no significant improvements in memory span and in the rhythm reproduction tasks. While these null findings are difficult to interpret, it may be that the specific rhythmic test used here was not best adapted to capture potential improvements or that training temporal processing may generalize to other cognitive functions without perceptible improvement on the trained function itself. Finally, correlational analyses between the different tasks did not reveal any significant results.

GENERAL DISCUSSION

Overall, our results provide convincing arguments in favor of using musical rehabilitative materials with children with dyslexia. The different aspects of the CMT program were specifically designed to improve sound perception, multiple aspects of temporal processing, and the integration of information from different sensory and motor modalities. In this respect, further experiments are needed to try disentangling the effects of these different components or, at least, to specify the weight of their respective contribution. Moreover, other characteristics of the training were: progressive learning, repetition of exercises, multiple modalities, and small-group workshops. Altogether, our results are in line with the repeated and longstanding observation from teachers, clinicians and scientists, that music in general, and perhaps more specifically learning an instrument, interfere positively with basic scholastic skills. Reading is the area that has been most directly tested probably as the most likely to have a direct impact on academic success. It was thus encouraging to find a significant impact of the CMT on reading ability.

The model most often put forward to account for a possible effect of music on cognitive development, specifically the acquisition of reading, calls upon a possible analogy between music and language. Many authors, most notably Patel (2003, 2011), have discussed this point, noting, in particular, that music and speech share many features such as the sequence of sounds, an alphabet that represents them, and a specific syntax. Patel (2011) hypothesized that music leads to adaptive brain plasticity of the same neural networks which are otherwise involved in language processing. More recently, it has been proposed to rather conceive music and language as sharing common cognitive resources, especially attentional and memory resources which could be equally recruited by music and language (Rogalsky et al., 2011; Perruchet and Poulin-Charronnat, 2013).

Concerning more specifically the topic of dyslexia, our results stand in favor of a role of music training in improving the

phonological deficit widely recognized as causal to the reading problems (Ramus, 2004), although others have questioned such an interpretation (Morais et al., 2010). In an influential theory of the mechanism underlying dyslexia, Goswami (Goswami et al., 2002, 2013) proposed that the dyslexics' cognitive system is specifically unable to process stimuli occurring at a frequency (frequency modulation) of the order of 2–10 cycles per second, which is the approximate frequency of syllables. As a consequence, dyslexics encounter difficulties capturing the segmental features of words and phrases. Thus, a developmental defect in processing the amplitude envelope of speech could lead to defective development of the phonological system in aspects related to the pace and patterns of intonation (prosody). In fact, children with dyslexia have been found to perform poorly on tasks of rhythmic perception and perception of musical meter (Huss et al., 2011). These observations have led researchers to propose rhythmic stimulation as a treatment for these aspects of the dyslexic deficit, and by extension, for the dyslexic disorder itself (Flaugnacco et al., 2015).

One of the strongest effect of the CMT program was on the attentional tests, in particular, the two subtests of the NEPSY auditory attention battery assessing selective and divided attention. An overall improvement of 20% over the 6 weeks of training is a successful outcome, in particular since this effect persisted after a 6 weeks untrained period. A recent brain imaging study (Heim et al., 2015) showed that dyslexics who received three different types of remediation (based on phonology, on attention, or on reading training) ultimately had similar pattern of improvement in terms of brain activation (specifically, an increase in activity in the lower left temporal region). It is thus conceivable that the improvements we have seen in our study on reading and phonology tasks are epiphenomenons, reflecting an impact on the attention system. We could not, however, find evidence of any correlations between the degree of improvement in phono-lexical tasks and attentional tasks. While the improvement was significant on tests of auditory attention, no significant changes were found on tests of visual attention, contrary to what one might expect if the CMT effect was operating through general attentional mechanisms. The fact remains that a positive effect on attention certainly contributed to the overall improvement even if the multi-faceted aspects of the CMT program preclude from concluding that this was the sole factor responsible for the improvements.

The cross-modal aspects of the CMT program may account for the effects found for tasks requiring sharing information between different modalities, such as reading and sequence comparison of letters as well as the phonological task, if one considers that such tasks require mandatory exchanges of information between the acoustic representation of phonemes that can be stored in auditory regions of the left temporal lobe, and the lower frontal areas, involved in phonological processing (Boets et al., 2013). Similarly, categorical perception also probably requires the involvement of structures such as the left frontal premotor areas that was found to be activated in dyslexics during a categorical perception task in a functional MRI experiment (Dufor et al., 2009). Finally, studies of brain plasticity in non musicians have shown that music training may

have the largest effects on brain anatomy and function when it combines sensory and motor training (Lappe et al., 2008, 2011). The recent literature on dyslexia and related learning deficits converge to show that the main structural differences in the brains of dyslexics compared to standard brains lie in the nature, integrity and directionality of certain hemispheric white matter bundles. These differences are present before learning to read (Saygin et al., 2013) and therefore can not be the result of a lack of experience with written language (although this may contribute as shown by studies of illiterates: Thiebaut de Schotten et al., 2014). These association bundles are also altered in musicians, both instrumentalists and singers (Halwani et al., 2011) and change in children after only a few months of musical training (Hyde et al., 2009). Although DTI is often regarded as a very indirect measure of white matter integrity (Jones et al., 2013), it remains that the same white matter bundles considered as the hallmark of the dyslexic brain are modified by musical training. Although strictly speculative, this observation deserves future investigation.

Using fMRI, Blau et al. (2009) have shown that dyslexics are characterized by poor integration of oral and written codes of the same phoneme. When these codes are congruent, temporal areas are less activated by combined visual-auditory processing than in controls; when the oral and written codes are incongruent, temporal areas are more strongly activated than in controls. An interpretation in terms of aberrant crossmodal integration can also potentially explain other clinical conditions such as dysgraphia, in which children hardly associate phonemes with their written form, or dyscalculia, in which they hardly relate numerical terms with the mental representation of the corresponding quantity (Noël et al., 2013).

Finally, we shall consider some of the potential caveats of the present study, specifically the absence of a control group, trained for comparison with another, already proven training method comparable in duration and cognitive load. Besides the obvious difficulty of finding such an ideal comparison group, our training protocol in Experiment 2 partly fulfills this objective, providing evidence for the selectivity of improvements, at least for some tasks, during the training period (T3 vs. T2) compared to the non-trained period (T2 vs. T1). Thus, potential motivational bias (so-called Hawthorne effect) can reasonably be ruled out by this design. Overall, we remain convinced that intra-subject approaches and disease progression models could be more convenient than larger samples for building comparison groups (since it is often difficult to recruit large samples in clinical settings). By having a pre-test/post-test design, we examined group improvement within subjects rather than between-subjects. With a lower number of individuals, this may have resulted in more extraneous effects due to individual differences. A second weakness is that improvement of specific cognitive mechanisms can not be distinguished from a purely attentional effect. In face of the consistency of improvement on attentional variables, the possibility of an exclusive or largely predominant impact of attention on executive processes can not be ruled out. Further experiments are needed to compare our results to the effect of a strictly attentional training protocol.

CONCLUSION

In a recent yet already acclaimed book, suggestively entitled “The Dyslexia Debate,” Elliott and Grigorenko (2014), two eminent specialists of the topic, provided arguments questioning the usefulness, or even reality, of the concept of dyslexia. This is based on the observation that various theories are proposed in the literature, none of them being entirely satisfactory and that most of the existing remediation methods are pedagogic rather than properly therapeutical. Accordingly, our conviction is that although the reality of a biological entity is indeed unquestionable, the multifaceted and kaleidoscopic clinical appearance of dyslexia, as suggested by the recent changes made to the DSM classification (APA, 2013), may lead to consider using multiple-component treatments, such as the ones offered by music training, rather than focusing on one single cognitive mechanism as in classical phonological training methods.

Music training may provide an ideal tool for such a new perspective: it allows considering each one of the multiple facets of dyslexia as a potential target to be improved. In this respect, music training may be one of the most complete and rational ways of treating dyslexia. Whatever the exact mechanism(s) subserving the observed improvements, their occurrence after relatively short sessions of musical training opens interesting avenues for future research as well as practical applications. First, our results suggest that several cognitive functions, including reading but not only, may be improved by adding a musical content to classical speech therapy and remediation of dyslexia.

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Musical plus phonological input for young foreign language readers

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Based on previous studies showing that phonological awareness is related to reading abilities and that music training improves phonological processing, the aim of the present study was to test for the efficiency of a new method for teaching to read in a foreign language. Specifically, we tested the efficacy of a phonological training program, with and without musical support that aimed at improving early reading skills in 7–8-year-old Spanish children ($n = 63$) learning English as a foreign language. Of interest was also to explore the impact of this training program on working memory and decoding skills. To achieve these goals we tested three groups of children before and after training: a control group, an experimental group with phonological non-musical intervention (active control), and an experimental group with musical intervention. Results clearly point to the beneficial effects of the phonological teaching approach but the further impact of the music support was not demonstrated. Moreover, while children in the music group showed low musical aptitudes before training, they nevertheless performed better than the control group. Therefore, the phonological training program with and without music support seem to have significant effects on early reading skills.

Keywords: phonological awareness, literacy, foreign language, reading, working memory, music

Introduction

A large amount of literature has been published on reading acquisition difficulties in native (L1) or in second language (L2) learning. Several factors, such as phonological and decoding skills have often been described as variables of crucial importance in the learning-to-read process (Brady, 1991; Melby-Lervåg et al., 2012). In their review, Hulme and Snowling's (2014) conclude that deficits in oral language skills as well as deficits in phonological language skills and problems in phoneme awareness, letter-sound knowledge and rapid automatized naming are of primary importance to account for learning to read difficulties. Jongejan et al. (2007) also considered that phonological language skills are important for L1 and L2 acquisition as they provide the necessary tools for lexical access and reading. The lack of oral language input in L2 acquisition is problematic when the pronunciation rules of L1 differ from L2. In this context, finding alternative research-based teaching approaches that could help learners to achieve foreign language literacy skills is very relevant.

Several results in the last two decades points to music as an aid in learning to read (Butzlaff, 2000; Bolduc, 2008; Standley, 2008; Lessard and Bolduc, 2011; Toscano-Fuentes and Fonseca-Mora, 2012) but the nature of this connection still needs to be clarified. Ott et al. (2011) suggest that early phonetic processing of verbal or non-verbal stimuli is differently organized depending on musical expertise. Patel (2011) proposes the OPERA hypothesis with 5 factors that may account for the influence of instrumental music training on brain plasticity and on shared

speech processing networks: Overlap in acoustic features in instrumental music and speech; Precision due to the higher demands of music; Emotion, Repetition and focused Attention. Christiner and Reiterer (2013) consider vocal music, singing, as a 'good indicator of the ability to remember new and unintelligible utterances' and conclude that the ability to sing improves auditory memory span. In their review of electrophysiological studies of speech segmentation, Schön and François (2011) conclude that musical expertise facilitates the learning of both linguistic and musical structures. Similarly, Schön et al. (2004) and Marques et al. (2007) demonstrate that musical training increased pitch discrimination in both music and language. Most importantly, children who are more sensitive in discriminating sounds due to music training are better on phonological awareness and reading tests (Lamb and Gregory, 1993; Douglas and Willats, 1994; Anvari et al., 2002; Peynircioglu et al., 2002; Bolduc and Montésinos-Gelet, 2005; Gromko, 2005; Forgeard et al., 2008; Moreno et al., 2009; Degé and Schwarzer, 2011; Herrera et al., 2011; Moritz et al., 2012).

Similarly, the four meta-analyses of Butzlaff (2000), Bolduc (2008), Standley (2008), and Lessard and Bolduc (2011) that reviewed more than 70 different multidisciplinary studies also point to a relationship between musical training and reading skills, mainly reading in L1. Butzlaff's meta-analysis reviewed 24 correlational and 6 experimental studies. The author concluded that results strongly and reliably associate music performance with standardized reading/verbal tests but that the causal nature of the relationship remained to be demonstrated. For instance, the influence of a factor such as teachers' expectancy could not be ruled out. Bolduc (2008) reviewed 13 studies and concluded that emergent literacy of preschoolers with or without learning difficulties is affected positively by musical instruction. Standley (2008) reviewed 30 studies related to music-related reading instruction and specific reading skills in order to make pedagogical recommendations about reading failure. The author differentiated two main music education styles underlying these studies: on the one hand, studies including multi-sensory programs based on Orff, Kodály, or Dalcroze methods that focus on singing, rhythm, instrument playing, or movement to music, and on the other hand, those that rely on extensive practice in choral, band, or orchestral ensembles. Although, the studies in general indicated benefits for reading, the great diversity of intervention programs and of variables such as age and motivation did not allow to draw firm conclusions, except that the younger the child, the stronger the gains from music interventions. According to this analysis, "Music activities that incorporate specific reading skills matched to the needs of children at-risk for reading difficulties (as well as special education, ESOL, early intervention) will enhance reading instruction" (Standley, 2008, p. 29). Finally, Lessard and Bolduc (2011) analyzed 17 studies that added evidence to the link between musical learning and reading among first to third graders. However, causality was not demonstrated due to differences between musical intervention programs, musical, and reading skills, sample sizes and also that many of these studies were unpublished works (doctoral dissertations, master thesis, pilot studies).

Turning to L2 acquisition, Fonseca-Mora and Gómez-Domínguez (in press) reviewed 27 experimental, correlational and quasi-experimental studies on music and language reading published between 2001 and 2013 and concluded that only 7.4% referred to L2 learning, thereby indicating a gap in this field. Marques et al. (2007) showed behavioral and electrophysiological evidence that musical expertise influenced the detection of pitch manipulations on sentence-final words in a foreign language. In this review, Chobert and Besson (2013) proposed that musical training may reduce phonological deficits in second language learning.

From an educational perspective, it remains unclear if the benefits for learning to read in L2 are based on general music instruction or on singing musically-supported phonological input matched to specific reading skills (Standley, 2008, p. 29). In a study with 11-year-old Spanish English Foreign Language (EFL) learners, Toscano-Fuentes and Fonseca-Mora (2012) showed that the use of musical-linguistic activities in the foreign language classroom improved reading skills as well as speaking and listening skills. Herrera et al. (2011) discussed the effects of a phonological and a musical plus phonological training program on the reading readiness of native and L2 Spanish-speaking children and stressed that the musical training approach helped native and foreign Spanish learners to outperform those without musical training in the ability to identify word endings, possibly because children's songs make rhyming words particularly salient (Herrera et al., 2011, p. 78). However, preschoolers who received the phonological training program without musical support obtained better results in phonological awareness and naming speed.

Our concern in this study is based on the fact that poor foreign language readers, in this case Spanish learners of English, lack phonological language skills, phoneme awareness, letter-sound knowledge and rapid automatized naming (Hulme and Snowling, 2014). In addition, the Spanish school curriculum does not include musical training. All second-grade Spanish children who participated in this study were very low-proficiency English language learners with classrooms located in suburban schools. This is important as this implies that there was no initial selection of participants. However, socio-cultural background, reading skills and working memory were assessed before training to ensure that the different groups were homogenous. Learners' musical aptitude was also tested as it has been described as an individual difference in language learning (Slevc and Miyake, 2006).

Purpose of the Current Study

Based on previous studies showing that phonological awareness is related to reading abilities and that music training improves phonological processing, the aim of the present study was to test for the efficiency of a new method for teaching to read in a foreign language. Specifically, we aimed at testing the efficacy of phonological training programs, with and without musical support that aimed at improving early reading skills in 7–8-year-old Spanish children learning English as a foreign language (EFL). Of interest was also to explore the impacts of these training programs on working memory and decoding skills. To achieve these goals we tested three groups of children: a control group, an experimental

group with non-musical intervention (active control), and an experimental group with musical intervention.

A video was selected in both experimental groups to teach early reading skills such as the alphabetic principle, phonological awareness and phonics. The musical experimental group was taught through video-clips that included musical elements such as songs with lyrics. The non-musical experimental group (or active control group) received the same phonological training program as the musical group but the program did not include melodies. The control group was taught in the traditional way without specific phonological awareness training nor musical support.

We hypothesized that the level of performance would be higher for teaching approaches that included phonological training with or without musical support than for traditional teaching methods. Moreover, we also hypothesized that musical support in a phonological training program for beginner EFL students would be an added value when learning to read because simple, rhythmic and repetitive melodies may induce the song-stuck-in-my-head phenomenon, a rehearsal loop that may improve sub-vocal rehearsal. The songs, created especially for improving phonetic aspects, were characterized by their slow pace and by the simplicity of their melodic contours. They were easy to memorize and, if activated periodically, they could favor automatized decoding. Finally, to determine the effects of the pedagogical intervention, pre/post tests and regression analyses including knowledge of sounds and letters, reading fluency and their interaction with working memory were computed.

Materials and Methods

Participants

A pre-post comparison design was used to examine training effects. Three second grade classes including 63 students ($\bar{X} = 7.6$ years old, $SD = 0.4$; 29 boys and 34 girls) were selected from two primary schools located in the same school district. Mean age between the three groups was not significantly different ($F < 1$) nor were the gender differences [$\chi^2_{(2, 63)} = 1.97, p = 0.374$]. At the beginning of the study, the music experimental group ($n = 18$) comprised 8 females and 10 males (mean age: $\bar{X} = 7.71$, $SD = 0.40$). The non-musical experimental group ($n = 22$) comprised 11 females and 11 males (mean age: $\bar{X} = 7.58$, $SD = 0.35$) and the control group ($n = 23$) comprised 15 females and 8 males (mean age: $\bar{X} = 7.67$, $SD = 0.51$).

Procedure

Prior to the beginning of the study, the school community was informed, organizational aspects were discussed and formal consent was granted. The control group and the non-music experimental group (with phonological training) were located within the same school. The music experimental group was located in a different school to avoid contamination if learners would sing the learned melodies in the playground. Teachers of both experimental groups were trained for several weeks before the start of the experiment. During the 2 weeks prior to the beginning of the training period, trained language graduate assistants and

graduate assistants in psychology (supervised by a neuropsychologist and two language researchers) tested the musical abilities, early reading skills, working memory and socio-cultural level of the 63 learners individually in a quiet room at their school. Immediately after the 11-week training period, reading skills and working memory of the young learners were tested again.

Questionnaires

A battery made up of four questionnaires was used:

- A socio-cultural survey, administered prior to the training program, to identify the main family characteristics and reading habits of the children.
- A musicality test to control for musical aptitudes. This test is an adaption of Hernández-Hernández and Santiago-González (2010) and included items that measured pitch, intensity, duration, rhythm, musical timbre and musical tempo. Two practice trials preceded each item to ensure that children understood the task.
- The *Wechsler Intelligence Scale for Children, 4th Edition* (WISC-IV, Spanish version) standardized neuropsychological assessment pre and post training. Selected tests included Digit Span and Letters and Numbers Sequencing subtests to assess auditory memory span.
- The *Early Grade Reading Assessment* (EGRA) in its English version including:
 - Letter name knowledge: name as many upper and lowercase letters as possible in 1 min. Letter presentation was random.
 - Initial sound identification: identify the initial sound of ten words read aloud by the test administrator.
 - Oral reading fluency: read a dialog with accuracy, speed and fluency in 1 min.

Training Program (Experimental Groups)

Children in both training programs received two 1-h sessions per week, for a period of 11 weeks and a total of 22 sessions. Video-clips were used in both training programs to help learners attach meaning to the minimal units of discursive articulation. Activities focused on the development of phonological awareness and phonics (e.g., auditory exercises that emphasized alliteration, word-onset awareness, and initial sound identification in frequent English words). Other activities focused on the learning of the alphabet (e.g., English letter-names and letter-sounds). The teacher in the music group used videos supported by songs with subtitles, characterized by simple and repetitive melodies and rhymes (Gértrudix Barrio and Gértrudix Barrio, 2010). Children in this group were trained in song perception and production and they were encouraged to sing the material learned in the hope that the catchy songs would foster self-initiated rehearsal. Children in the non-musical group worked on the same reading skills and contents but through attractive and colorful videos, posters, and audio-books without music. Both teachers planned together and simultaneously their lessons so that they were teaching the same thematic units at the same time.

Phonological Training Program

The phonological training program included the following tasks that were supported by visual materials (e.g., posters and flashcards):

Practicing with Single-Letter Sounds

Children learned the names and sounds of the letters of the English alphabet. Letter-names and letter-sounds were presented using videos, posters, and audio-books in the non-musical group, and using songs and subtitles the music group.

To establish a relationship between letters and sounds, frequent one-to-three syllable words were spelled and pronounced at the same time the songs and non-musical videos were played. Every word with a common spelling and phonetic pattern was classified into different word-bank lists written on a board. For example, words with the same middle sound as “book” (/u/ sound: *book, foot, look, food*) or those with the “ph” grapheme (/f/ sound: *elephant, phone, dolphin*) were included in the same list. This task was used for students to automatize graphemes-phonemes matching as well as English pronunciation, structures, and rules. Most of the tasks were designed to foster learners to use their auditory discrimination and production skills, such as:

(a) Onset and rime detection tasks: learners were asked to identify initial and final phonemes in words. For example, “What is the first sound in the word ‘fish’? or ‘What is the last sound in ‘fox’?”

Phonological oddity tasks were also included in which learners were asked to spot the odd word out when listening to three different words, two of them sharing the same initial phoneme (e.g., “which word begins with a different sound: jam, yoghurt, juice?”).

(b) Oral blending skills, manipulation of sounds in words and word formation tasks when learning and reading new words. Learners were required to change the initial sound of a word to create a new word. For example, to change the initial sound of the following words (*hen, hill, hat, hot, hump*) to /p/ or to choose which words could be made with the following initial phonemes: c, b, l, f, v, h, j. Tasks that required learners to change the middle vowel in a word to another that had the same sound to find out the correct spelling that matched a picture presented on a flashcard (“jamper or jumper?”, “mauth or mouth?”, “food or fud?”).

Phonics and Spelling

Word choice tasks based on spelling were also instructed: learners made words using various combinations of vowel and consonant letter-cards, putting them on a board for all students to see. Sound matching tasks based on blending words onset graphemes and ending phonemes (rimes), using “the phonic wheel,” were to improve learner’s spelling skills.

Traditional Program (Control Group)

The traditional teaching program was based on the idea that phonological decoding skills are learned from direct exposure to foreign language and transfer directly from L1. The teacher used the syllabic and global word approach as classically described in L1 textbooks. The curriculum for teaching English to second

graders mainly included vocabulary (numbers, colors, food, animals, parts of the house, verbs), some easy verbal routines (greeting, saying good-bye...) and simple sentences such as “I have/not..., I like/ I don’t like...” Flashcards and games were used to help students to increase motivation for the English lesson.

Data Analysis

One-Way repeated measures Analyses of Variance (ANOVAs) were conducted to test for before training differences in socio-cultural factors and musical aptitude between groups. Moreover, ANOVAs were also computed to test for differences before and after training that included Group (Control, musical, and non-musical) as well as Session (pre vs. post training) as factors. Finally, multiple regression analyses with interactions (Aiken and West, 1991; Rosel et al., 2014) were also conducted to test for the effects of the training program in the experimental and control groups. Data analysis was performed using the 21.0 SPSS statistics package.

To determine whether the intervention produces different effects in the 3 groups (non-musical experimental, musical experimental, and control groups), an ANCOVA was conducted on the differences between groups after training, controlling for the level of performance before training (i.e., “prior knowledge”) for each one of the three tasks. In addition, we computed a regression analysis for each one of the three tasks. Reading variables_{pre}, WM_{pre} and the interactions with the Group factor (musical, non-musical, and control) were included.

Results

Before Training

The three groups were homogeneous regarding their socio-cultural background (see **Table 1**). Learners’ musical aptitude was also homogeneous within the three groups before training, with a normal distribution [Kolmogorov-Smirnov’s test: K-S = 0.622, $p = 0.834$ and Levene_(2, 60) = 0.495, $p = 0.612$]. However, results revealed significant differences between groups [$F_{(2, 60)} = 14.175$, $p < 0.001$]: mean scores in the non-musical experimental (NME) group ($\bar{X} = 27.2$, $Sd = 3.7$) were significantly higher than in the control group (Cont: $\bar{X} = 22.6$, $Sd = 3.2$;

TABLE 1 | Test for between groups differences on socio-cultural variables.

	<i>df</i>	<i>X</i> ²	<i>P</i>
Level of studies (father)	10	10.22	0.42
Level of studies (mother)	10	7.67	0.66
Same language spoken at home and at school	2	0.83	0.66
Home language other than Spanish	2	3.16	0.20
Reading at home besides schoolwork	2	0.79	0.67
Family member who reads more	2	3.99	0.13
Someone reading aloud to participants	2	3.10	0.21
Listening to music	2	0.20	0.90
Frequency of listening to music	6	3.73	0.71

Bonferroni = -4.62 , $p < 0.01$) and in the musical experimental group (ME: $\bar{X} = 21.3$, $Sd = 4.4$; Bonferroni = -5.89 , $p < 0.01$), with no significant differences between the control and musical group (Bonferroni = 1.27 , $p < 0.855$).

Working memory (WM) data showed a normal distribution ($K-S = 0.495$, $p = 0.967$), but this was not the case for “Correct Letters read in English” ($K-S = 4.069$, $p < 0.001$), “Initial Sound Identification” ($K-S = 1.021$, $p = 0.021$) and “Correct Words Read in a Dialog in English” ($K-S = 3.659$, $p < 0.001$). Non-significant differences were found between the three groups [$F_{(2, 60)} = 0.55$, $p = 0.58$]. The H non-parametric test of Kruskal-Wallis showed no between-groups differences in the “Correct letters read in English” ($H_{K-W(2)} = 2.977$, $p = 0.226$) and the “Correct words read in a dialog in English” ($H_{K-W(2)} = 5.159$, $p = 0.076$) tasks, but significant differences in the “Initial sound identification” task ($H_{K-W(2)} = 6.562$, $p = 0.038$), with higher scores in ME than in NME group ($Md_M = 94$, $Md_{N-M} = 100$, 5 ; $U_{M-W} = 110$, $p = 0.016$).

In sum, the three groups were similar in terms of socio-cultural background and working memory but the non-musical experimental group had significantly higher musical aptitudes than the other two groups and the musical experimental group showed higher scores in the “Initial sound identification” task than the NME group.

Before vs. after Training Comparisons

Results of non-parametric Wilcoxon tests for the variables with non-normal distribution were all significant: the level of performance in “Correct letters read in English” ($Z_W = 4.791$, $p. 001$), “Correct words read in English dialogs” ($Z_W = 3.429$, $p. 001$) and “Initial sound identification” ($Z_W = 3.679$, $p. 001$) were higher after than before training. By contrast, results for WM (ANOVAs) were not significant [$F_{(2, 61)} = 0.001$, $p = 0.974$] and neither was the interaction between Session (WM_{pre} vs. WM_{post}) and Group [ME, NME and Cont; $F_{(2, 62)} = 1.90$, $p = 0.16$]. Thus, we decided to use the results obtained for WM_{pre} to avoid the potential influence of the WISC test on WM_{post}.

“Correct Letter Names Read in English” Task

The main effect of Group was significant after training [$F_{(2, 60)} = 9.81$, $p < 0.01$, $\eta_p^2 = 0.247$] with a larger effect when “prior knowledge” was controlled for [$F_{(2, 59)} = 16.16$, $p < 0.01$, $\eta_p^2 = 0.354$, $\beta = 0.979$], explaining 41.5% of the variance ($R_c^2 = 0.415$). Planned Bonferroni contrasts revealed that the level of performance increased significantly in both experimental groups compared to the control group ($p < 0.01$, IC_{EM-C} [5.11, 16.56], IC_{ENM-C} [5.67, 16.44], with no differences between the musical and non-musical experimental groups ($p > 0.05$, IC_{EM-ENM} [−5.95, 5.50]).

Table 2 shows the interaction terms between working memory scores in the control group and in the non-musical group through dummy variables, with the musical experimental group as reference. Specifically, the model explained 52.2% variance in number of letters read per minute ($R^2 = 0.52$). The number of correct letters read was predicted by the combined effect of Group and WM, with significantly lower scores in the control group than in the musical group.

TABLE 2 | Regression coefficients for “correct letters read in English” task after training.

	Unstdndzed Coeff	Std. error	t	Sig.
(Constant)	−20,600	11,026	−1868	0.067
Combined punctuation in WM _{pre}	0.326	0.115	2832	0.006
Correct letters read _{pre}	0.563	0.126	4477	0.000
Dummy non-musical experimental	22,919	13,671	1676	0.099
Dummy control	22,364	13,436	1665	0.102
Interaction WM _{pre} —Dummy non-musical group (Ref. G. M. Exp.)	−0.243	0.141	−1728	0.090
Interaction WM _{pre} —Dummy control group (Ref. G. M. Exp.)	−0.351	0.141	−2484	0.016

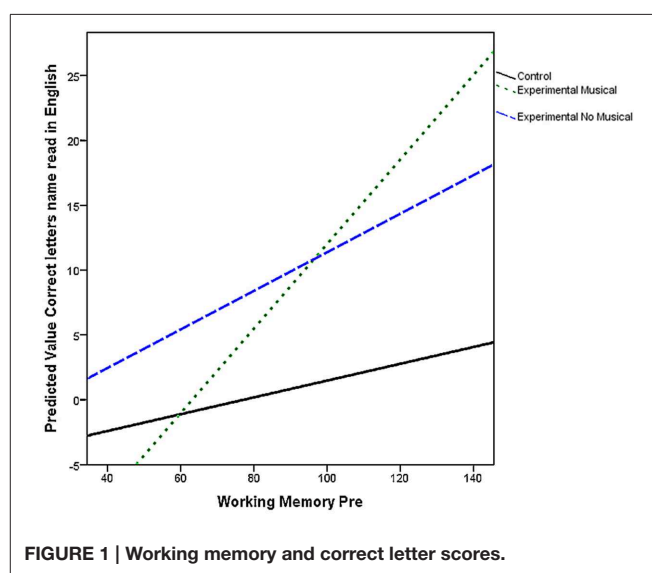


FIGURE 1 | Working memory and correct letter scores.

As can be seen on **Figure 1**, learners with higher WM_{pre} scores before training improved more in this task with larger improvements in the musical group. After training, the difference between the musical and control group was significant with no difference between the experimental (non-musical and musical) groups.

Correct Words Read in a Dialog in English

The main effect of Group was significant after the intervention [$F_{(2, 60)} = 5.216$, $p = 0.008$, $\eta_p^2 = 0.148$, $\beta = 0.812$]. However, no pre-post differences were found when “prior knowledge” was controlled for [$F_{(2, 59)} = 1.018$, $p = 0.368$, $\eta_p^2 = 0.034$]. The model explained 65.6% of the variance ($R_c^2 = 0.656$) with confidence intervals at 95% (Bonferroni tests: IC_{CTR-EM} [−3.22, 5.18], $IC_{CTR-ENM}$ [−5.31, 2.36], IC_{EM-ENM} [−6.82, 1.90]).

As can be seen on **Table 3**, scores obtained in both groups, NME group (lower initial value) and Cont group (higher initial value), were based on significantly different initial values than in the ME group. Although, the interaction was not significant, there was a trend for the largest increase in this task to be found

TABLE 3 | Regression Coefficients for “correct words read in English” post.

	Unstandardized Coefficients	Std. error	T	Sig.
(Constant)	−8.57	5.69	−1.50	0.14
Combined punctuation in WM _{pre}	0.18	0.06	3.23	0.00
Correct words read in a dialog in English _{pre}	−0.01	0.01	−0.87	0.39
Dummy non-musical experimental	7.48	2.53	2.96	0.00
Dummy control	5.29	2.49	2.12	0.04

in the NME group (steeper slope), then in the ME group and the slowest evolution to be found in the Cont group. In this case, $R^2 = 0.505$ is reached.

It can be observed (**Figure 2**) that the non-musical group (lower initial score) and the control group (higher initial score) have significantly different initial values from the musical group. Differences were observed in the intercepts (the value of Y when X = 0).

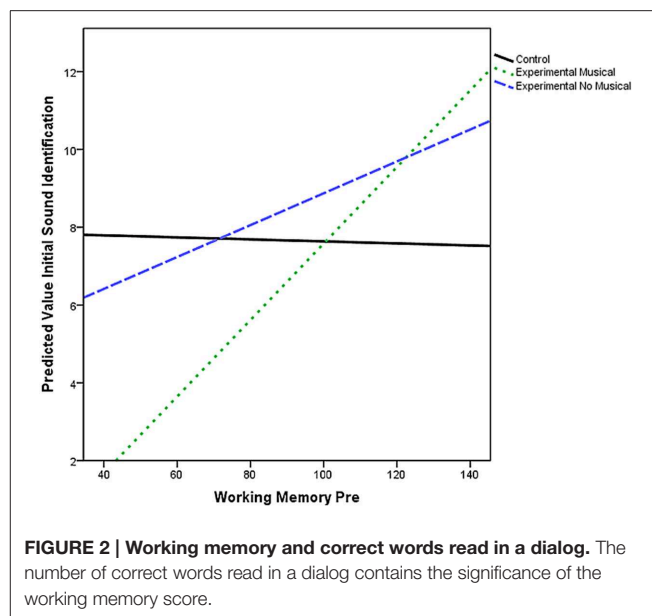
Initial Sound Identification

After training, the main effect of Group was significant [$F_{(2, 60)} = 3.352$, $p = 0.042$, $\eta_p^2 = 0.101$, $\beta = 0.612$]. However, no pre-post differences were found when “prior knowledge” was controlled for [$F_{(2, 59)} = 1.47$, $p = 0.602$, $\eta_p^2 = 0.017$]. The model explained 45.3% of the variance ($R_c^2 = 0.453$) with confidence intervals at 95% (Bonferroni tests: IC_{CTR-EM} [−1.30, 1.36], IC_{CTR-ENM} [−1.74, 0.81], IC_{EM-ENM} [−1.90, 0.91]). Significance of the interaction terms between working memory and control group scores contrasted with the musical group on the total scores in this task is reported in **Table 4**. In this model $R^2 = 0.525$ is reached.

As can be seen on **Figure 3**, no improvement was found in the control group. By contrast, the level of performance improved in both experimental groups with higher scores in the musical group. The difference between control and musical group was significant with no difference between the musical and non-musical experimental groups.

Discussion

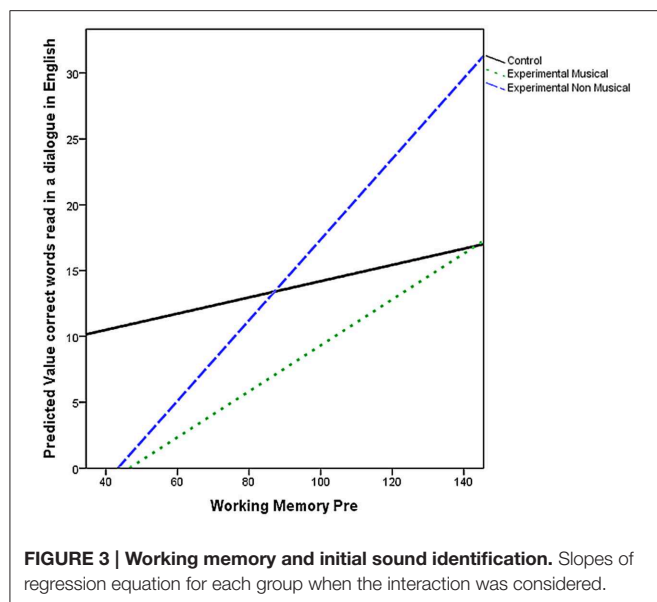
The main objective of this study was to examine the effects of phonological training programs with and without music support on reading abilities in 7–8 year-old Spanish children learning English as a foreign language. A positive outcome would allow us to propose an alternative, research-based, foreign language teaching method. Most studies point to instrumental musical training as an important factor contributing to reading skills (Anvari et al., 2002; Peynircioglu et al., 2002; Bolduc and Montésinos-Gelet, 2005; Gromko, 2005; Forgeard et al., 2008; Moreno et al., 2009; Degé and Schwarzer, 2011; Herrera et al., 2011; Moritz et al., 2012). However, instrumental musical training is often difficult to implement in primary schools when music classes are not included in the curricula. By contrast, singing is often

**FIGURE 2 | Working memory and correct words read in a dialog.** The number of correct words read in a dialog contains the significance of the working memory score.**TABLE 4 | Regression Coefficients for “initial sound identification in English” post.**

	Unstdndized Coeffs.	Std. error	t	Sig.
(Constant)	−1.16	2.65	−0.44	0.66
Combined punctuation in WM _{pre}	0.06	0.03	1.99	0.05
Initial sound identification _{pre}	0.50	0.09	5.71	0.00
Dummy non-musical experimental	5.21	3.28	1.59	0.12
Dummy control	7.36	3.24	2.274	0.03
Interaction WM _{pre} —Dummy non-musical group (Ref. G. M. Exp)	−0.05	0.03	−1.466	0.15
Interaction WM _{pre} —Dummy control group (Ref. G. M. Exp)	−0.08	0.03	−2.289	0.03

practiced in kindergarten and early primary schools. Christiner and Reiterer (2013) pointed out that singing is similar to music at the acoustic-perception level and can also help detecting rhythmic cues in foreign languages. Consequently, singing can contribute to improve speech production and is easier to implement. Thus, rather receiving an instrumental music training, the young foreign language learners involved in this experiment benefitted from a phonological training program based on repeatedly singing rhythmic melodies during the 11-weeks training program.

Results of pre vs. post comparisons showed that children in the musical and non-musical (i.e., active control) training groups performed significantly better than children in the control group regarding the “Correct letters read in English” and the “Initial sound identification” tests. Moreover, a trend was found in the “Correct words read in an English dialog” with larger increase in the non-musical group compared to the musical group and



smallest increase in the control group. Finally, predictive analyses based on regressions with interactions, and taking working memory into account, indicated no significant differences between musical and non-musical groups, both doing better than the control group. These results clearly point to the beneficial effects of the phonological teaching approach but the further impact of the music support was not demonstrated.

Children in the non-musical group performed higher than children in the musical and control groups in the musicality test presented before training. This was possibly linked to these children being from Spanish gypsy families who typically show strong rhythmic abilities (Gil and Azcune, 2012). In this respect, using a longitudinal approach, David et al. (2007) showed that sensitivity to musical rhythm was related to the ability of decoding complex words requiring the use of linguistic stress and they argued that rhythm predicted reading ability from grade 1 to 5 in primary school. Similarly, Moritz et al. (2012) concluded that the rhythmic abilities students developed when they were preschoolers correlated with their phonological abilities in second grade. Thus, the higher level of performance of children in the non-musical group in both the “correct letters read in English” and the “correct words read in an English dialog” may be linked to English being a stress-timed language (while Spanish is syllable-timed) strongly relying on rhythmic cues. Moreover, early phonetic processing may be organized differently in children with high musical aptitudes, as shown in adults with stronger musical expertise (Ott et al., 2011). In other words, learners with higher musical aptitudes may tend to benefit more from the phonological training program than children with lower musical aptitudes. Importantly, however, while children in the music group showed low musical aptitudes before training, they nevertheless performed better than the control group in the tests described above. Therefore, the phonological training programs with and without music support seem to have significant effects on early reading skills.

Of interest in this study was to examine the influence of working memory and how it interacted with the effects of other factors. Previous reports in the literature have shown that instrumental musical training significantly improved working memory (Ho et al., 2003; Franklin et al., 2008). More recently, Christiner and Reiterer (2013) showed that singing also improved auditory working memory span in Austrian adult singers performing in Hindi. Surprisingly, no such differences between WM_{pre} and WM_{post} were found in the present study. However, as seen in **Figure 1**, the largest increase in WM scores is found in the music group. Moreover, the higher the scores in WM_{pre} , the larger the increase in “correct letters read” scores after training. This effect was larger in the experimental groups than in the control group with no differences between experimental groups. It may be that the speech sounds as well as the visual and orthographical elements, addressing the phonological loop and the visuo-spatial sketchpad, respectively (Baddeley, 2012), used in the two experimental groups, improved learners’ basic reading skills. In addition, and in line with Christiner and Reiterer (2013) results, it may be that the repetitive use of melodies with memorable lyrics allowed young learners to better retain verbal materials and relevant foreign speech sounds. Finally, the lowest level of performance in the control group clearly showed that direct transfer from learners’ L1 reading skills to another language should not be taken as granted in the foreign language classroom.

One final aspect that deserves comments is the positive impact of the phonological and musical-phonological programs after a relatively short training duration (11 weeks). This finding is in line with previous results showing significant effects of training after 16, 14, and 4 weeks, respectively in Gromko (2005), Moreno and Besson (2006), and Register et al. (2007). In addition, our results are in line with the conclusions from a meta-analysis conducted by Standley (2008) showing that using music activities that matched the specific reading needs of the children was more important than the duration of training (e.g., training of less than 4 weeks ($d = 0.61$) were equally effective than training over an entire school year (0.33 , $p = 0.37$). Nevertheless, and in line with previous longitudinal studies over a school year or longer (David et al., 2007; Moreno et al., 2009; Moritz et al., 2012; Chobert et al., 2014), it would be of interest in further experiments to test for the effects of the phonological and phonological plus music training programs on foreign learning abilities when these programs are applied for a longer duration.

Conclusions

Acquiring good phonological and decoding skills is of uttermost importance for foreign language learners and these abilities are not necessarily directly transferred from L1 knowledge (specifically when L1 and L2 rely on different phonological systems). Nevertheless, these abilities are needed to access lexical content while reading. The phonological training program based on visual support that was used in this study, improved some of the early reading skills in 7–8-year-old Spanish EFL students. Moreover, learners in

the phonological plus musical support training program outperformed children in the control group. Thus, simple rhythmic melodies that work as carriers of visual and orthographic perception may stimulate the rehearsal needed for improving specific phonological skills, thereby providing valuable teaching approaches for learning to read in a foreign language.

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Engagement in community music classes sparks neuroplasticity and language development in children from disadvantaged backgrounds

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Children from disadvantaged backgrounds often face impoverished auditory environments, such as greater exposure to ambient noise and fewer opportunities to participate in complex language interactions during development. These circumstances increase their risk for academic failure and dropout. Given the academic and neural benefits associated with musicianship, music training may be one method for providing auditory enrichment to children from disadvantaged backgrounds. We followed a group of primary-school students from gang reduction zones in Los Angeles, CA, USA for 2 years as they participated in Harmony Project. By providing free community music instruction for disadvantaged children, Harmony Project promotes the healthy development of children as learners, the development of children as ambassadors of peace and understanding, and the development of stronger communities. Children who were more engaged in the music program—as defined by better attendance and classroom participation—developed stronger brain encoding of speech after 2 years than their less-engaged peers in the program. Additionally, children who were more engaged in the program showed increases in reading scores, while those less engaged did not show improvements. The neural gains accompanying music engagement were seen in the very measures of neural speech processing that are weaker in children from disadvantaged backgrounds. Our results suggest that community music programs such as Harmony Project provide a form of auditory enrichment that counteracts some of the biological adversities of growing up in poverty, and can further support community-based interventions aimed at improving child health and wellness.

Keywords: low socioeconomic status/poverty, community music training, electrophysiology, reading, speech, auditory training

INTRODUCTION

Over 16 million children in the US live in families with incomes below the federal poverty level, with a disproportionate number (68%) being members of minority racial and ethnic groups (Jiang et al., 2014). Parental income, occupation, and education are commonly combined to define a child's socio-economic status [SES (Sirin, 2005)]. A child's SES can interact with other personal factors (race, ethnicity, gender, etc.) to lead to weaker academic achievement (Bradley and Corwyn, 2002; Sirin, 2005; Hernandez, 2011; Skoe et al., 2013) and lower high school graduation rates (Ensminger and Slusarcick, 1992; Bradley and Corwyn, 2002), with lower academic achievement persisting even for low SES students who attend college (Walpole, 2003).

Auditory processing skills, known to be important for language development (Benasich et al., 2002, 2008; Boets et al., 2011; Goswami et al., 2011), may contribute to the link between SES and academic achievement. Children from low SES backgrounds have greater daily exposure to noise (Adler and Newman, 2002;

Evans and Kantrowitz, 2002) and tend to be less concerned with using hearing protection in excessively noisy contexts like concerts (Vogel et al., 2007). Chronic noise exposure in children has been linked to weaker reading proficiency and cognitive skills (Maxwell and Evans, 2000; Haines et al., 2001; Clark et al., 2005) and can lead to delayed auditory neural development and greater spontaneous neural activity in animals (Chang and Merzenich, 2003; Seki and Eggermont, 2003; Zhu et al., 2014). Additionally, children from low SES backgrounds hear less complex language and fewer words overall during early language development, which contributes to weaker vocabularies when entering school (Bradley and Corwyn, 2002; Hoff, 2003; Cartmill et al., 2013). Similarly, a host of neural systems important for language, memory, and cognition are impacted by SES background (Raizada et al., 2008; Hackman et al., 2010; Noble et al., 2012), including those implicated in auditory attention (D'Angiulli et al., 2008; Stevens et al., 2009) and speech encoding (Skoe et al., 2013). Encouragingly though, community and home based interventions may reverse these effects; children from low SES families participating in Head Start and attention

training with their parents showed improved language, cognition, and neural measures of auditory attention relative to children in Head Start programs alone (Neville et al., 2013).

Musical training is another avenue of enrichment that may counteract some of the auditory deprivation endemic to low SES environments. A number of studies have revealed that children undergoing music training have stronger cognitive abilities, vocabulary, rhythm perception and production (linked to reading skill), perception of vocal pitch, and perception of speech in noisy backgrounds than non-musician children (Ho et al., 2003; Schellenberg, 2004, 2006; Magne et al., 2006; Forgeard et al., 2008b; Hyde et al., 2009; Moreno et al., 2009, 2011; Strait et al., 2011; Dege and Schwarzer, 2012; Strait et al., 2012; Slater et al., 2013; Chobert et al., 2014; Seither-Preisler et al., 2014; Strait and Kraus, 2014). Additionally, musical practice can strengthen children's auditory encoding of speech (Magne et al., 2006; Besson et al., 2007; Chobert et al., 2011; Strait et al., 2011, 2013; Tierney et al., 2013; Chobert et al., 2014; see Strait and Kraus, 2014 for a review), auditory discrimination and attention (Koelsch et al., 2003; Moreno et al., 2009; Chobert et al., 2011; Putkinen et al., 2013), and lead to structural changes in auditory cortical areas (Hyde et al., 2009; Seither-Preisler et al., 2014). The auditory benefits of music training have direct implications for language skills and academic achievement (Hetland and Winner, 2010; Corrigan and Trainor, 2011; see Tierney and Kraus, 2013 for a review); accordingly, music may serve as an effective training tool for children with learning and attention impairments (Overy, 2003; Bhide et al., 2013; Seither-Preisler et al., 2014).

Community music programs, such as El Sistema and Harmony Project, provide students from low SES backgrounds with music opportunities that enrich the students and their communities. El Sistema, founded almost 40 years ago, provides over 500,000 Venezuelan children with free musical training in their community (for a review of the program, see Majno, 2012). Children are commonly enrolled as young as 2 or 3 years old and are supported through their teen years. Graduates of the program commonly return to teach at their community music center and parents are continually educated by the organization on how to support and encourage their child's music as they advance. Since 2009, 62 El Sistema inspired music programs have been started in the United States. Harmony Project (Los Angeles, CA, USA) similarly promotes the development of healthy children and communities by providing free music training to children from low SES backgrounds in gang-reduction zones of Los Angeles, CA, USA (www.harmony-project.org). Children learn basic music skills in a preparatory class, eventually receive a free instrument to participate in group classes, and have opportunities for performance and ensemble playing throughout their enrollment from early grade school to high school. Between 2010 and 2014, 93% of Harmony Project alumni enrolled in post-secondary education, versus 67.6% of students graduating from public schools within Los Angeles County [most recent data from California Department of Education: Educational Demographics Unit (2008–2009) and Harmony Project Whole Notes 2013–2014 (2014)].

Our laboratory has shown that participation in music training through Harmony Project can reinforce literacy skills, enhance

the perception of speech in background noise, and strengthen the neural encoding of speech sounds in children from low SES backgrounds (Kraus et al., 2014a,b; Slater et al., 2014a,b; Kraus and Strait, in press). Here we explore how the extent of student *engagement* in instrumental classes mediates music training's effects on neural speech processing. Although we do not employ an active control group, our aim is to show *within a group undergoing musical training* how engagement may influence the benefits seen for students. Music engagement was defined by a student's percent attendance in class and teacher ratings of classroom participation. We investigated whether greater engagement in music instruction can positively impact the neural encoding of speech. We focused on measures of subcortical brain activity reflecting speech harmonics, the consistency of the response, and spontaneous neural activity, all of which are weaker in teens from low SES backgrounds relative to higher-SES peers (Skoe et al., 2013).

MATERIALS AND METHODS

PARTICIPANTS

Twenty-six children participated in the study (13 males, ages 6 years 11 months – 9 years 3 months; $M = 8$ years 5 months). All children were public elementary school students living in the gang reduction zones of Los Angeles, CA, USA. Participants attended schools where $\geq 90\%$ of students qualify for free or reduced lunch. Families qualify for free or reduced lunch when their income is below 185% of the federal poverty level (U.S. Department of Education, 2014). The average education of the participants' mothers was 10.7 years ($SD = 4.2$) and the median and modal maternal educational attainment level was completion of high school/GED. Maternal education is one of the strongest predictors of SES (Hoff et al., 2012), and 76% of children whose parents have a high school degree or less live in low-income families (Jiang et al., 2014). Average maternal education was equivalent across the four Harmony Project sites in which students were enrolled (Kruskal Wallis Test: $\chi^2 = 6.252$, $p = 0.100$).

Participants were excluded if they had a history of neurological disorders, IQ less than 80 [Wechsler Abbreviated Scale of Intelligence (Woerner and Overstreet, 1999)], previous musical training, or failed a hearing screening (air-conduction thresholds above 20 dB HL for octaves 125–8000 Hz). All procedures were approved by the Northwestern University Institutional Review Board and informed parental consent was obtained for all participants.

MUSIC INSTRUCTION

Students were followed as they participated in Harmony Project for 2 years. The project curriculum started students in music appreciation class, where they learned pitch-matching and rhythm skills, musical styles and notation, and basic vocal performance and recorder playing. Participants attended these classes twice weekly for 3–10 months ($M = 5$).

As musical instruments became available and students were judged to be ready, they progressed to instrumental instruction. Students were given their own instruments and participated in a mix of group-based instrumental classes for approximately 4 h per week. Students in this study participated in one of four Harmony Project sites. The description of weekly classes at each site

and the number of students at each site is presented in **Table 1**. Students played a number of different instruments (viola, cello, bass, French horn, clarinet, or trumpet) and the number of students playing each type of instrument is also reported in **Table 1**. Harmony Project has a standard curriculum for each instrument type (woodwinds, brass, and strings), including rigorous mastery benchmarks required for advancement to the next level of the curriculum. For example, Level 1 benchmarks require students to: demonstrate basic knowledge of music concepts such as rhythm and intonation, exhibit instrument specific skills and knowledge such as proper posture and correct clef identification, and present evidence of commitment through practice at home. As students progress, expectations shift to more instrument specific skills (e.g., proper bowing or breathing techniques) and knowledge and use of dynamics, key signatures and deviations, and articulations (e.g., staccato). Students are required to learn to read music and play from memory throughout the program. All students in the study transitioned to instrument use during their first year, so at the end of their second year in Harmony Project, students had an average of 165 h of school-based instrumental training (SD = 40) and over 210 h of music training overall.

Teacher ratings of music engagement

At the end of each instrumental class, teachers reported the percent attendance for each student (hours attended/total hours possible) and rated the students on their level of participation in class (1-not at all, 2-little, 3-moderate, 4-fairly good, 5-complete). We averaged percent attendance and teacher ratings of class participation across all the instrumental classes taken by each child over the 2 years. Both attendance and teacher perception of student effort predict ‘behavioral engagement’ in school, reflecting a student’s involvement in the participatory aspects of school (Glanville and Wildhagen, 2007). Because neither percent attendance nor participation were significantly correlated with the number of instrumental classes taken (p -values > 0.310), we are confident that higher levels of class participation or attendance reflect student motivation and are not simply artifacts of being enrolled in more classes. Additionally, percent attendance

and participation ratings did not differ across Harmony Project sites (Kruskal Wallis Test: $\chi^2 = 5.366$, $p = 0.147$; $\chi^2 = 4.781$, $p = 0.189$, respectively), suggesting no rating bias among the sites.

READING ASSESSMENT

Before enrolling in Harmony Project and after 2 years of Harmony participation, students were administered the Test of Oral Word Reading Efficiency (TOWRE; Torgesen et al., 1999). The TOWRE, a measure of reading fluency, comprises word and non-word reading subtests. Children are required to read a list aloud as quickly as possible and the number of words (or non-words) read correctly in 45 s is tallied and combined to form the Total score.

NEUROPHYSIOLOGICAL MEASURES

Evoked potentials were collected from the auditory brain-stem in response to a 40-ms synthesized speech syllable [da], using an Intelligent Hearing Systems SmartEP system equipped with a cABR module (Miami, FL, USA). Stimuli were presented to the right ear at 80 dB SPL at a rate of 10.9 Hz through electromagnetically-shielded earphones (ER-3A, Etymotic Research, Elk Grove Village, IL, USA). Responses to opposing polarities were subtracted and filtered online from 0.1 to 1.5 kHz in the test session before music training began and from 0.05 to 3 kHz in the test session after 2 years of music training. Due to the differences in filter settings, which were expanded as the longitudinal project evolved, within-subject comparisons over the two test dates are not possible.

Neural measures were those previously shown to index SES (Skoe et al., 2013). Measures of response consistency, speech harmonics, and spontaneous neural activity were generated in MATLAB (Mathworks, Natick, MA, USA). Response consistency reflects the replicability of the response over the first half and second half of the recording on a scale from –1 to 1. Values were Fisher-transformed for analyses, but Pearson’s r -values are shown in **Figures 1** and **2**. Speech harmonics comprise the average amplitude of the frequency following response over the first-formant range of the stimulus

Table 1 | Student instrument experience by site and by instrument.

Harmony Project site	Typical class participation	Number of students by instrument type
Alexandria elementary school	–1 h instrumental class twice per week –2 h string ensemble rehearsal weekly	4 (bass)
Beyond the bell	–2 h ensemble rehearsals twice per week (includes pull-out sectional rehearsals)	12 (2 clarinet, 3 flute, 7 trumpet)
EXPO enter (YOLA)	–1 h instrumental class each week –3 h ensemble rehearsal weekly	5 (2 cello, 1 French horn, 1 viola, 1 trumpet)
Hollywood	–1 h instrumental class twice per week –3 h ensemble rehearsal (concert band) weekly	5 (trumpet)

Participants were enrolled in music classes in one of four sites and played one of seven different instruments. Details about the standard class schedule of each site is included.

(264–656 Hz). Spontaneous neural activity reflects the amplitude of brainstem activity in the absence of the stimulus, and likely reflects neural noise. Please see Banai et al. (2009), Hornickel and Kraus (2013), and Skoe et al. (2013) for additional details.

STATISTICAL ANALYSES

The relationships among instrumental class attendance, class participation, and neural measures were evaluated at each of the two test dates using Spearman's correlations due to the non-normal distributions of the music engagement variables. Spearman's correlations are more conservative than Pearson's correlations because they reduce the impact of outlying data points and are most appropriate for smaller sample sizes. Similar to Pearson's correlations, Spearman's rho is a

direct measure of effect size, reflecting the proportion of variance in the dependent variable (neural measures) accounted for by the independent variable (music engagement variables). Correlation coefficients with magnitudes of 0.1 are considered small effect sizes, 0.3 medium, and 0.5 large (Cohen, 1992). Reading fluency scores were compared using a paired *t*-test.

RESULTS

Children who had better attendance in instrumental music classes over 2 years had stronger neural encoding of speech harmonics and better response consistency, two measures previously linked to SES (see **Figure 1**; **Table 2A**). Additionally, there was a positive association between class participation and response consistency (see **Figure 2** for two representative participants

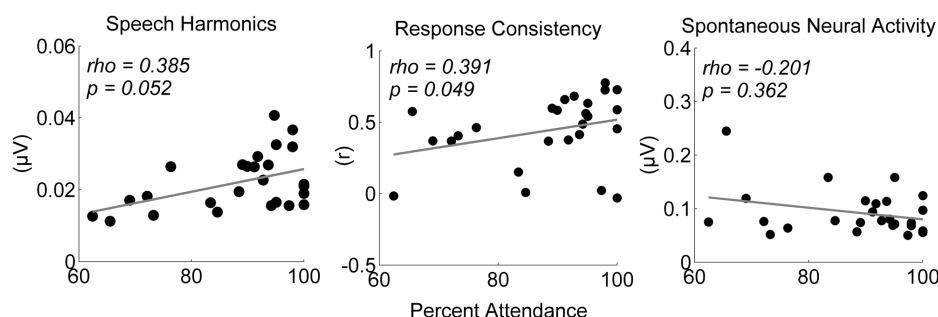


FIGURE 1 | Children who regularly attended instrumental classes had stronger neural encoding of speech after 2 years, particularly for measures of speech harmonics and response consistency.

Neural measures before music training began did not predict attendance or level of class participation, suggesting greater engagement in music classes may lead to stronger neural encoding

of speech and not vice versa. Speech Harmonics is measured as the average amplitude of harmonic encoding in the frequency following response (μV), Response Consistency is measured as the Pearson's correlation coefficient between response replications (*r*), and Spontaneous Neural Activity is measured as the root-mean-square magnitude of the pre-stimulus (μV).

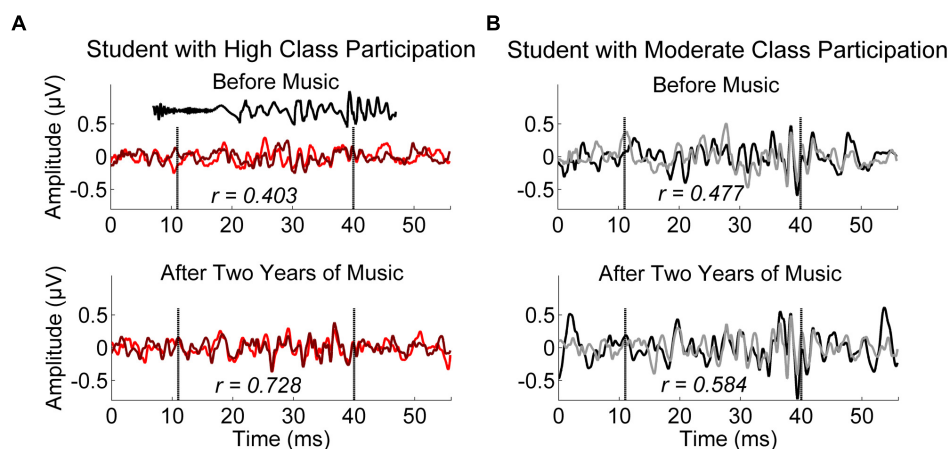


FIGURE 2 | Children who were most engaged during instrument classes had more consistent neural responses to speech after 2 years.

(A) (red/maroon), a representative subject who had “complete” participation as rated by multiple teachers. The [da] stimulus is plotted in black in top panel of A for reference, shifted in time to account for neural delay. (B) (black/gray), a representative subject who had “moderate” participation as rated by multiple teachers. Both students completed four instrumental classes over

the course of 2 years. Before music training, (top) the participants do not differ greatly in the consistency of their neural response to speech. After 2 years of music training (bottom), however, the child who participated more in class has a more consistent response to speech than the child who participated less. The two traces in each panel represent the two replications of the response, collected as the recording procedure dictates. The time region of the analysis is marked with vertical hashed lines in each panel.

and **Table 2A**). Interestingly, the least engaged participants in the study still had “moderate” class participation, suggesting that even small variations in student engagement can have implications for success with music training. No relationship was found between music engagement and spontaneous neural activity.

Importantly, our results suggest that greater engagement in music classes predicts stronger speech encoding, not vice versa. Neural measures before music training did not predict subsequent attendance or class participation (p -values > 0.126, see **Table 2B**), which suggests that participants with strong speech encoding to begin with did not necessarily go on to be the most engaged students in the study. Additionally, the small variation in years of maternal education did not predict attendance or class participation ($\rho = -0.302, p = 0.142; \rho = 0.126, p = 0.548$).

Participants overall showed a subclinical yet statistically-significant decline in reading fluency over the 2 years [$M1(SD1) = 109.27(12.76), M2(SD2) = 106.04(15.22), t_{25} = 2.456, p = 0.021$]. However, change in reading fluency scores was strongly correlated with engagement in instrumental classes. Children who participated more in class were more likely to show an improvement in reading fluency, while those who participated less were more likely to show a decrease in reading fluency ($\rho = 0.443, p = 0.023$).

DISCUSSION

Here we found that greater engagement in group music instruction by children from low SES backgrounds predicted stronger neural encoding of speech for measures negatively influenced by low SES. Children who attended class more regularly and had better classroom participation had stronger neural encoding of speech after 2 years of music training than did their less-engaged peers. These results were found *within* a group of children undergoing music training, and without an active control group. It is likely that greater engagement in other extracurricular activities could also yield stronger neural outcomes. Perhaps the novel experience of

participating in an extracurricular activity, interacting with the researchers, etc., influenced our findings. However, previous studies that have employed active control groups indicate that music training but not art training strengthens auditory skills (Moreno et al., 2009, 2011). Moreover, child musicians differ from non-musician children on auditory working memory and attention but not visual analogs (Strait et al., 2014). Similar to our analyses within a group of children participating in music, Hyde et al. (2009) found that the quality of musical training is critical for neuroplasticity. Children participating in private keyboard lessons for 15 months showed structural growth in primary auditory cortex, correlated with their improvement in melody and rhythm perception tasks; however, a second group of children who participated in school music classes each week learning basic singing and drumming skills did not show the same neural growth (Hyde et al., 2009). Our results suggest that engagement is an important factor mediating the benefits seen from musical training. It’s important to note that we found these relationships between engagement and neural function within a group of students whose attendance was relatively high on average (88%), who were rated as having ‘moderate’ or better class participation, and whose families were highly motivated for them to participate. The limited variance in music engagement and probable influence of other factors such as personality on student engagement may have contributed to the modest statistical significance of our tests. Nevertheless, we do see moderately strong relationships between engagement and neural measures, suggesting that even if well-motivated to begin music training, students may not make gains unless they are actively engaged in the process. In the same vein, previous studies of musicians have revealed the important role of continued practice in maintaining the benefits of musicianship (Pantev et al., 1998; Ho et al., 2003; Norton et al., 2005; Forgeard et al., 2008b; see Strait and Kraus, 2014 for a review).

Greater attendance and class participation were not predicted by neural measures before musical training. This suggests that children with the strongest neural function before beginning musical practice did not go on to be those most engaged in music classes. Instead, it appears that greater engagement in music may have resulted in stronger neural encoding. Cohort research revealing neural differences between musicians and non-musicians is unable to definitively say whether those differences were pre-existing, although the oft observed relationship between length of practice and benefits of musicianship supports music experience as the cause (Pantev et al., 1998; Ho et al., 2003; Norton et al., 2005; Forgeard et al., 2008b; Seither-Preisler et al., 2014; see Strait and Kraus, 2014 for a review). A host of recent longitudinal studies report that musical training can selectively enhance auditory function in children without pre-existing differences. When children are randomly assigned to music or art training, only those in musical training show enhancements in auditory neurophysiology and attention (Moreno et al., 2009, 2011; Chobert et al., 2014). Additionally, children who elect to participate in music do not have inherent differences in neural structure or function from their peers (Norton et al., 2005; Hyde et al., 2009; Tierney et al., 2013), but show enhancements in auditory system structure and function after 1–2 years of musical training (Hyde et al., 2009; Tierney et al., 2013; Kraus et al., 2014b). We were not able to conduct paired

Table 2 | (A) Children who are more engaged in music classes have stronger neural encoding of speech after music training for measures previously linked to SES (speech harmonics and response consistency). **(B)** Neural measures before beginning musical training do not predict subsequent engagement in music classes.

	Music engagement	
	Percent attendance	Class participation
A		
Speech harmonics	0.385 (0.052)	0.242 (0.235)
Response consistency	0.391 (0.048)	0.389 (0.049)
Spontaneous neural activity	−0.201 (0.326)	0.013 (0.949)
B		
Speech harmonics	−0.150 (0.464)	0.119 (0.562)
Response consistency	0.280 (0.166)	0.138 (0.502)
Spontaneous neural activity	−0.308 (0.126)	−0.212 (0.299)

Values are Spearman’s rho (p -value). Significant relationships are bolded.

pre-post comparisons due to alterations to our recording scheme as the study progressed. However, the lack of relationship between pre-test neural measures and subsequent classroom engagement in our study suggests our results are independent of pre-existing differences in neural function.

Although some studies report no differences in cognition for children who elect to participate in music versus those who do not (Schlaug et al., 2005; Hyde et al., 2009), personality and pre-existing skill may nevertheless influence a student's engagement and persistence in music. For example, using duration of music lessons as the outcome variable, Corrigan et al. (2013) showed that cognitive skills (along with parental income) predicted how long students continued with music. They additionally showed that children exhibiting high Openness to experience (part of the Big Five personality traits) were more likely to continue with music lessons after taking into account cognition and parental income (Corrigan et al., 2013). In our group of students, personality factors may have influenced attendance and participation in music classes. Those who were highly engaged in music classes may have been highly engaged throughout their school day, had a more encouraging home environment, may have been more motivated individuals, etc. Indeed, Openness as a personality trait is linked to factors reflecting academic engagement and achievement in college students, such as enjoying thinking and analyzing, enjoying connecting with others in class, and enjoying working hard (Komarraju and Karau, 2005). One limitation of our study is the lack of measures to investigate which of these personality factors may predict engagement in music. Importantly though, duration of musical experience also predicts cognition and language skills (Schellenberg, 2006; Forgeard et al., 2008b), suggesting the presence of a reinforcing loop. Children more open to new experiences may elect to begin music lessons and continue to participate longer, which leads to larger cognitive benefits, predicting continued retention in music, etc.

In addition to enhancing cognitive skills, music training can lead to stronger reading, language, and academic skills in children (Schellenberg, 2004, 2006; Hetland and Winner, 2010; Corrigan and Trainor, 2011; Dege and Schwarzer, 2012; Francois et al., 2013; Seither-Preisler et al., 2014; Slater et al., 2014a; see Tierney and Kraus, 2013 for a review), including those with reading impairments (Overy, 2003; Bhide et al., 2013). As a group our participants showed a small decline in reading fluency scores, which likely reflects a trend for the achievement gap to widen for children from low SES backgrounds as they age (Sirin, 2005). However, the change in reading scores varied with music engagement; reading fluency improved in children with greater class participation. That this relationship was seen despite the small variance in class participation ratings speaks to the importance of active engagement in music for engendering benefits. Students who had the highest class participation rankings were more likely to show an increase in reading score after 2 years of participation in Harmony Project. On the other hand, participants who had 'moderate' class participation on average were likely to show a decrease in reading score. As the consistency of the auditory brainstem response to speech and the strength of representations of the speech harmonics have been repeatedly linked to reading ability (Banai et al., 2009; Hornickel et al., 2011, 2012a; Hornickel and Kraus, 2013; White-Schwoch and

Kraus, 2013), and musical aptitude can predict reading ability (Ho et al., 2003; Forgeard et al., 2008a; Huss et al., 2011; Strait et al., 2011), it is possible that stronger speech encoding arising from greater participation in music classes contributed to improved reading scores.

Animal models of auditory deprivation and enrichment yield insight into the potential mechanisms involved in the neuroplasticity observed here. Similar to humans exposed to chronic noise, animals reared in noisy environments show detrimental effects on auditory processing, such as broader neural frequency selectivity (Zhou et al., 2011; Zhu et al., 2014), greater spontaneous neural activity (Seki and Eggermont, 2003), and weaker neural speech processing (Reed et al., 2014). Additionally, rats with a knock-down of a dyslexia-linked gene show more variable cortical responses to speech and weaker neural differentiation of speech (Centanni et al., 2014a). These deficits are thought to be due to imbalances in excitatory and inhibitory neurotransmitters, also shown to be negatively affected by noise exposure during development (Guo et al., 2012; Zhu et al., 2014). Auditory enrichment and behavioral training can reverse these effects, yielding better neural frequency selectivity, more consistent neural responses to speech, better neural differentiation of speech sounds, and can re-establish excitatory/inhibitory balances in rats with early auditory deprivation (Guo et al., 2012; Centanni et al., 2014b; Zhu et al., 2014). Although these exact mechanisms have not been investigated in humans, it is very likely that similar ones are at play. Like the auditory enrichment paradigms in animal studies, music training is a social, multi-sensory process that activates multiple neural networks during repeated exposure to the enriching "environment" [please see Patel (2011) for a review of the multi-faceted nature of music training].

We failed to find significant relationships between spontaneous neural activity and musical engagement. Previous studies of auditory training in humans, such as music training, computer-based training games, foreign language learning, and classroom acoustic modification, have shown improvements in the evoked response to speech, but none have reported changes in spontaneous neural activity (Besson et al., 2007; Song et al., 2008; Stevens et al., 2008; Russo et al., 2010; Hornickel et al., 2012b; Strait et al., 2013). Animals that are subjected to noise during development have increased spontaneous neural activity, likely due to decreased neural inhibition as a result of abnormal excitatory/inhibitory neurotransmitter ratios (Seki and Eggermont, 2003; Zhu et al., 2014). However, auditory enrichment can alleviate imbalances in excitatory and inhibitory neurotransmitters (Guo et al., 2012; Zhu et al., 2014) and possibly reduce noise-induced spontaneous neural activity. Spontaneous cortical activity thought to reflect a functional "resting-state network" can be altered with training in humans (Lewis et al., 2009; Taubert et al., 2011). The present findings show that the extent of music engagement tracks with changes in stimulus-evoked, but not spontaneous background activity; training-related influences on spontaneous activity should be explored further.

Our results support the importance of active experience and meaningful engagement with sound to engender neural changes.

Even in a group of highly motivated students, small variations in music engagement (attendance and class participation) predicted the strength of speech encoding after music training. These measures of neural encoding are known to be weaker in children from low SES backgrounds. Although personality features likely played a large role in students' levels of engagement in class, our results do support that greater levels of engagement are not prompted by pre-existing differences in neural encoding. In our group of participants from low SES backgrounds, those who were the most engaged in music classes had the strongest neural responses and were most likely to show improvements in reading ability. Community music programs such as Harmony Project and El Sistema have proven success in bolstering academic achievement in children from disadvantaged backgrounds. We have also shown that participation in Harmony Project reinforces literacy skills and enhances the neural encoding of speech cues important for reading and the perception of speech in noisy backgrounds (Kraus et al., 2014a,b; Slater et al., 2014a; Kraus and Strait, in press). Taken together, we suggest that motivated engagement in community music programs may counteract some of the auditory impoverishment that children from low SES backgrounds commonly experience.

AUTHOR CONTRIBUTIONS

Nina Kraus and Dana L. Strait designed the study; Dana L. Strait, Jessica Slater, and Elaine Thompson collected the data; Jane Hornickel analyzed the data; Jane Hornickel and Nina Kraus prepared the manuscript; all authors contributed to the final version of the manuscript.

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A groundwork for allostatic neuro-education

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We propose to enliven educational practice by marrying a conception of education as guided human development, to an advanced scientific understanding of the brain known as allostasis (stability through change). The result is a groundwork for allostatic neuro-education (GANE). Education as development encompasses practices including the organic (homeschooling and related traditions), cognitive acquisition (emphasis on standards and testing), and the constructivist (aimed to support adaptive creativity for both learner and society). Allostasis views change to be the norm in biology, defines success in contexts of complex natural environments rather than controlled settings, and identifies the brain as the organ of central command. Allostatic neuro-education contrasts with education focused dominantly on testing, or neuroscience based on homeostasis (stability through constancy). The GANE perspective is to view learners in terms of their neurodevelopmental trajectories; its objective is to support authentic freedom, mediated by competent, integrated, and expansive executive functionality (concordant with the philosophy of freedom of Rudolf Steiner); and its strategy is to be attuned to rhythms in various forms (including those of autonomic arousal described in polyvagal theory) so as to enable experiential excitement for learning. The GANE presents a variety of testable hypotheses, and studies that explore prevention or mitigation of the effects of early life adversity or toxic stress on learning and development may be of particular importance. Case studies are presented illustrating use of allostatic neurotechnology by an adolescent male carrying diagnoses of Asperger's syndrome and attention-deficit hyperactivity disorder, and a grade school girl with reading difficulties. The GANE is intended as a re-visioning of education that may serve both learners and society to be better prepared for the accelerating changes of the 21st century.

Keywords: allostasis, neuro-education, neurodevelopment, RDoC, executive function, polyvagal theory, toxic stress, neurotechnology

Introduction and Orientation

One should not ask, 'What does a person need to know and be able to do for the existing social order?' but rather, 'What gifts does a person possess and how may these be developed?' Then it will be possible to bring to society new forces from each succeeding generation. Then the social order will always be alive with that which fully developed individuals bring with them into life, rather than that each succeeding generation be made to conform to an existing social organization – Rudolf Steiner, Founder of Waldorf Schools.

Fenner and Rapisardo (1999)

Today, clinicians are accused of overdiagnosing and overmedicating children with behavioral problems. It is time to shift from an exclusive focus on behavior and symptom-based diagnosis to incorporate a deeper understanding of neurodevelopmental trajectories with interventions that can support the healthy development of brain and behavior – Thomas Insel, Director of the US National Institute of Mental Health.

Insel (2014)

Educational practice is as old as human civilization. Modern understanding of the brain, supported by innovations in technology, has developed over approximately the last 100 years. The proposition of this paper is that a recent innovation in scientific understanding of the brain known as *allostasis* (“stability through change”) has potential to support a constructive re-visioning of education as stewardship of emergent, individualized, and advanced brain functionality. The progeny of the union between education and allostasis is an approach to the developing and multi-faceted learner that we call allostatic neuro-education. To characterize this conceptualization we outline a perspective, an objective, and a strategy for educators which collectively constitute a *groundwork for allostatic neuro-education* (GANE). The ultimate benefit we conceive from the GANE is to support the profession of education to reach its own highest potential, to be a match for the meaning reflected in its Latin etymology *educere*, to *lead out* the expression of that which is within. To support understanding of the terms, concepts, and abbreviations presented in this paper, a glossary is provided in **Figure 1**.

For purposes of introducing the reconstructed view of education entailed by the GANE, we recapitulate a taxonomy outlined by Lawrence Kohlberg, the pioneer of moral psychology, and Rochelle Mayer in a paper titled “Development as the Aim of Education” (Kohlberg and Mayer, 1972). In this work, Kohlberg and Mayer categorized educational practice in the Western world to consist of three broadly different streams or typologies – the romantic, the transmission of culture, and the progressive. To update their labels, we substitute the words *organic* for their *romantic*, *cognitive acquisition* for their *cultural transmission*, and *constructivist* for their *progressive*.

The *organic* type of educational practice is intended to nurture that which is already within the child. Cognitive development is encouraged (rather than instructed) to proceed along lines of pre-programmed unfolding and in parallel with physical and emotional development. Organic education is philosophically descended from Jean Jacques Rousseau (1712–1778) and others who exalt the natural and express concern over the corrupting influences of society. Formal and didactic aspects of education tend to be de-emphasized in favor of child-centric practices including homeschooling, de-schooling, and other strategies aimed to enable flourishing of innate potential. Education for *cognitive acquisition* is rooted in classical traditions of Western civilization, aiming to impart literacy, mathematical competence, and other forms of knowledge that relate to prevalent norms. Education for cognitive acquisition is largely society-centric, and it is largely synonymous with education as instruction (from the Latin *instruere*, to build up or pile on) that includes emphases on standardized testing. In the USA, coalescence

around standards of cognitive skill acquisition has occurred in various ways including federal legislation known as No Child Left Behind as well as the more recent initiative known as the Common Core. In East Asian cultures, cognitive acquisition takes on even greater importance in that educational striving is commonly used as a tool for character building or as a high-stakes funnel for social advancement (Chua, 2011; Ripley, 2014). *Constructivist* education, represented by John Dewey (1859–1952) and others, posits that the goal of education is to facilitate guided development of the learner through proactive experiences and interactions with society and the natural environment at large, such that knowledge and skills are actively created through engagement with living and changing contexts. Dewey in particular considered that education to nurture such development should have implications for both the learner and for healthy participatory democracy.

Kohlberg and Mayer associated the first two educational streams with different metaphors for learning. Organic education compares the learner to a plant or blossoming flower. For education in the service of cognitive acquisition, the learner has inputs and outputs, comparable to a machine or other functional instrument. For the constructivist, the learner is understood to be engaged in a constant dialectic with the environment. She or he generates and applies an initial idea, experiences its consequences, and subsequently revises the idea or its application in a continuous procession. Kohlberg and Mayer were proponents of constructivism, proposing that this approach was the only one adequate to “prepare free people for factual and moral choices which they will inevitably confront in society.” Their sentiment is echoed in other recent writing that has warned against the societal impacts of educational systems that begin and terminate with unbridled focus on cognitive acquisition. Such approaches can fail to recognize fundamental misalignments between learner needs and teacher efforts, ultimately resulting in wasted resources and distorted understanding, akin to “lecturing birds how to fly” (Taleb, 2012).

The GANE in contrast recognizes important roles for all three educational streams or traditions, and the dialectical appreciation that Kohlberg and Mayer assigned exclusively to the constructivist can be understood to apply to all three traditions aligned in a larger unfolding. The child-centric view of organic education can be understood, normatively, as a robust starting point for the learner, as a way to nurture the innate capacities of a young life form. The society-centric view of education for cognitive acquisition may serve to equip and ballast developing learners with a common set of shared skills and knowledge which enable clear and robust communal interaction, without which self-centered individuals may descend into narcissism or chaos. Though constructivism may represent the highest educational ideal, in its goal of enabling perpetual renewal and re-creation for both individual and society, actual constructivist practice may need a foundation of nurturing, experience, skills, and knowledge that are the province of organic and cognitive acquisitional approaches. The GANE thus aims for a re-visioning of education that is inclusive of a breadth of traditions.

The GANE is motivated by a sense of urgency for educators to consider seriously the need for innovation in educational

organic education – learning as pre-programmed unfolding of innate potential. May use metaphor of the learner as a blossoming plant, and may be associated with “de-schooling” or homeschooling. Referred to by Kohlberg and Mayer (1972) as “romantic.”

cognitive acquisitional education – learning as acquisition of common and core skills and information. May imply metaphor of the learner as machine or a functional instrument. Typically associated with standardized achievement tests. Referred to by Kohlberg and Mayer as “cultural transmission.”

constructivist education – learning as development of actively created skills and knowledge that emerge at the guided interface between the learner and the environment, intended to enable adaptive creativity and perpetual renewal for both the individual and society. Referred to by Kohlberg and Mayer as “progressive.”

homeostasis – stability through constancy, a paradigm for understanding physiological regulation that defines life as being preserved through defense against change. Originated in controlled setting of laboratory-based experimentation on animals, prior to the dissemination and acceptance of evolutionary theory. Interventions intend to modify functionality toward a level deemed normal and normative, and/or to correct functioning of mechanisms in the direction of the defined norm.

allostasis – stability through change, a paradigm for physiological regulation consistent with evolutionary theory, that recognizes change, not constancy, as the norm; defines success as fitness in the context of complex natural contexts; and identifies the brain as the organ of central command. Originated in observations regarding psychosocial and environmental influences on health. Interventions aim to facilitate responsiveness to the full range of signals from many sources.

education – in context of allostasis, activity intended to educe (lead out, from the Latin *educere*) the expression of innate potential; in context of homeostasis, activity intended to instruct (to build up or pile, from the Latin *instruere*) or otherwise aid cognitive acquisition in the direction of a standardized norm.

allostatic neuro-education – activity intended to educe the full potentiality of learners, with respect for their integrated neurodevelopmental trajectories, to support successful engagement with complex and unpredictable environments. Encompasses organic, cognitive acquisitional, and constructivist practices.

GANÉ – groundwork for allostatic neuro-education, consisting of a perspective that encompasses long-range neurodevelopmental trajectories, an objective of authentic freedom mediated by competent, integrated, and expansive executive functionality, and a strategy of attending to rhythms, especially rhythms of arousal that permit experiential excitement for learning.

neurodevelopmental trajectory – unique growth path for a neural system, subsystem, or collection of systems, subject to alteration by use or environmental exposures over an extended time frame.

toxic stress – strong, frequent, or prolonged activation of the body’s stress response systems, in the absence of the buffering protection of a supportive relationship.

RDoC – Research Domain Criteria. Framework put forth by the US National Institutes of Mental Health in 2009, to advance mental and behavioral health science by grounding understandings in brain biology, instead of reliance on symptom and behavior-based diagnostic categories. Preliminarily, cognitive systems, arousal, positive valence, negative valence, and social systems are recognized as basic domains of brain function that cut across disorders and have a continuum of expression from health through disease.

freedom – in context of allostatic neuro-education, a conjoint state of mind and brain reflecting advanced executive functionality and maximal optionality, requiring symphonic activity in multiple brain domains. In the philosophy of Rudolf Steiner (1894), a state characterized by use of moral imagination permitting ethical individualism, for evolutionary advance of both individual and society.

mental health – as defined by Sterling (2014), responsiveness of the conscious and unconscious mind to the full range of signals from many sources; the capacity to choose among thoughts and shift flexibly between them, to match mood and affective expression in ways optimal for the current or anticipated context. (Cf mental disorder – reduced capacity for mentation, emotions or behaviors to shift in the service of responsive adaptation or anticipation; being “stuck”).

polyvagal theory – hierarchical model for understanding functionality of the autonomic nervous system. Recognizes two different forms of parasympathetic low arousal. Capacity for self-calming (myelinated vagus nerve) represents an evolutionarily recent form of self-regulation, whereas a freeze mode (unmyelinated vagus) is associated with attention to novelty or experience of a significant stressor, and is derived from evolutionarily ancient anatomy. Capacity for mobilization, or sympathetic fight/flight, is hierarchically positioned between self-calming (most advanced mode) and the freeze state. In the context of allostatic neuro-education, attention to rhythmic shift between fine-tuned parasympathetic self-calming and sympathetic arousal represents a strategy for learning and engagement, by supporting alternation between interoceptive consideration and excitement for learning, respectively.

allostatic neurotechnology – any technology designed to respect and support the brain as organ of central command, without presuming to define normative set points for operation or to intervene directly on brain mechanisms.

FIGURE 1 | Glossary of terms, concepts, and abbreviations presented in the paper, listed in the order in which they are principally discussed in the text.

principles and practice. Because of the acceleration of changes that we are now experiencing in multiple realms – socio-economic, cultural, geo-political, and technological – educational systems must better prepare 21st century learners to be adaptable and genuinely constructivist, so they may thrive in a world that is manifesting increasing degrees of variability and unpredictability. To further explicate the background of

the GANÉ and how it differs from other approaches to neuro-education, the ensuing section of the paper introduces the advanced model of physiological regulation known as *allostasis*. Allostasis is proposed as a necessary advance on the paradigm of homeostasis, that has limitations associated with its origins in pre-evolutionary 19th century biology. The third section defines and characterizes the GANÉ by outlining its perspective,

objective, and strategy. Fourth, the paper considers a range of testable hypotheses that stem from the GANE. To emphasize that the brain-centrism of the GANE is not metaphorical or only implied, two case studies are presented that illustrate use of allostatic neurotechnology by learners with special needs.

Homeostasis, Allostasis, and Education

The advanced understanding of brain functionality that is central to the GANE is contained in the paradigm of *allostasis* (Sterling and Eyer, 1988), defined as “stability through change.” In comparison to the idea of homeostasis, or “stability through constancy,” allostasis presents a more biologically accurate understanding of the role and functionality of the brain itself (Sterling, 2004, 2012). There are three main insights from the paradigm of allostasis that enable it to serve as a robust scientific foundation for neuro-education. First, it recognizes that change and not constancy is the fundamental character of biological regulation. Second, it views life and the definition of success against the background of complex natural contexts rather than controlled settings. Third, allostasis identifies the brain as the organ of central command.

The innovative view of allostasis may be better understood by an overview of the paradigm it aims to subsume. The foundations of modern experimental physiology were laid by Claude Bernard (1813–1878) and Walter Cannon (1871–1945) through the use of laboratory-based animal experimentation methods and reasoning based on reductive materialism. Bernard posited that physiological systems are designed to preserve a constant interior environment – that is to say, that the goal of life forms is to defend their internal biological operations against changes wrought by the environment. Central to the paradigm was the presumption that differences between species were not of great significance, and thus inferences about the functioning of physiological systems drawn from experiments on one species could be applied to others. Homeostasis recognizes the individual as a collection of multiple and distinct systems – the heart and circulatory system, the gut and digestive system, and others – and that illumination of the operative mechanisms of these systems is the basis for therapeutics.

In contrast, the paradigm of allostasis was born from recognition that social and environmental variables have critical influence on biological functioning, and that life is not fundamentally oriented toward “defense” of a constant internal environment (Sterling, 2004, 2012). Rather, in alignment with evolutionary theory, biological systems are understood to function in such way that they normatively change their operational set points to be optimally fit for the context of the natural environment, which is essentially complex, variable, and unpredictable. *Under evolutionary theory, the ultimate goal of life is not to defend against changes but rather to create more life*, and this objective is achieved through adaptation to present and anticipated needs. Critically, allostasis recognizes that to coordinate and adjudicate among the competing demands of different organ systems or functionalities, life requires a higher-order faculty for organization and regulation, and the organ for

such faculty is the brain. The brain is the seat of central command, guiding the organism as a unit to adapt to new circumstances, and to prepare for changes yet to come.

Biological systems under conditions of environmental monotony (including the controlled settings of a laboratory) may appear to be homeostatic because their set points are not challenged to need modification. Nonetheless homeostasis and allostasis present different understandings of the underlying drivers of physiological regulation and their implications. Homeostasis implies the existence of normal and normative set points, whereas allostasis does not (Sterling, 2012, 2014). Under allostasis, recognition that brain activity is fundamentally context-dependent is critical to understanding health or disease across systems, whereas under homeostasis the focus is on identifying disturbances in molecular mechanisms. Homeostatic modeling invites strategies to modify mechanisms and their set points toward the putative norms. Allostatic modeling recognizes the need for variability in biological expression and specifically aims to support the brain’s native ingenuity for central command (Sterling, 2004, 2014). Notably it is not the anatomical focus *per se* of a particular strategy that makes it homeostatic or allostatic, and interventions directed toward the brain can be of either type.

Dominance of the homeostatic paradigm in physiology and biomedical health care can be considered the neuroscientific analog of the dominance of standardized cognitive acquisition as the mode of educational practice in the modern era. Just as the modern biomedical system views the individual as so many distinct organ systems, so does educational practice for cognitive acquisition view the learner in terms of a collection of different and distinct systems for cognitive, emotional, and behavioral functionalities, and others. As with homeostatic reductionism, these systems are held to have independent mechanisms of operation, and in general only the cognitive is under the purview of the secular educator. Other systems are allocated to an array of specialists– psychologists, psychiatrists, other behavioral specialists, physical education instructors, speech or language therapists, music or art specialists, and others. Modern educators are largely not trained to interact with learners as whole and integrated beings, and even less are they trained to consider that the learner’s various domains may have a common upstream source of regulation in the brain.

The allostatic educator is dynamically flexible and uses different educational approaches in ways that fit the need. During the earliest phases of life, there is major biological development in neural systems that may be well supported by organic educational practices. Education for cognitive acquisition or cultural transmission supports learners to “stand on the shoulders of giants” with respect to established questions. In its essence, allostasis is especially aligned with constructivist educational practices – when learners are ready to question the question itself – which are attuned to the contingent and changing nature of skills and knowledge. By the lights of allostatic neuro-education, the most well educated student is not the one with the highest scores on fixed and standardized tests, but the one who is most robustly positioned to create successful interactions with complex, variable, and unpredictable environments. Capacity

for such success has evolutionary consequences for both the individual and society.

At this juncture we consider whether the proposition to use allostasis as the basis for a new vision of education represents a form of the naturalistic fallacy. That is to say, though change may be the norm of nature, and though the brain's biological role may be to serve as the organ of central command, it may not necessarily be warranted to presume that educators should use these concepts as normative guidelines for educational practice. For example, biological evolutionary theory has been used notoriously, and fallaciously, to justify policies that are intrinsically political (and often repressive), including social darwinism and eugenics. To this question, the authors answer (with further discussion in the following section) that the GANE has an explicitly philosophical dimension, and it specifically aims for attainment of a state of advanced executive functionality associated with authentic *freedom*. Freedom is represented by maximal *optionality* for both the learner and educator. Allostatic neuro-education is based on the power to *opt*, not the imperative of the *ought*. Optionality includes the freedom to place a relative emphasis on cognitive acquisitional educational practices that are themselves essentially homeostatic. It is rather the homeostatic paradigm that tends to commit the naturalistic fallacy, in conceiving that the constant (or typically average) set points for biological regulation should be considered normative, and therefore defended by means of therapeutic or otherwise normalizing interventions.

Perspective, Objective, and Strategy of the GANE

We define allostatic neuro-education as *activity intended to educe (lead out) the full potentiality of learners, with respect for the integrated developmental trajectory of their multi-faceted brains, to support their successful engagement with complex, changing, and unpredictable environments*. Allostatic neuro-education may derive, now or in the future, from a range of principles and practices in education, child development, neuroscience, brain-focused technologies, or other fields. In this section we give initial substance to the GANE under the rubric of a perspective, an objective, and a strategy.

The GANE Perspective is the Learner's Extended Neurodevelopmental Trajectory

In bringing a developmental perspective to the foreground, the GANE aligns with pedagogical strategies introduced by a series of educators and psychologists beginning in the late 19th century. Rudolf Steiner (1861–1925) founded the Waldorf School in 1919, based on an appreciation of multiple domains of the human being that develop over the course of 7-years periods. Waldorf teachers progress with the same students from year to year so they can carefully steward the child's long-term process for completing these tasks, and educational practices are designed to encompass organic, cognitive acquisitional, and constructivist objectives. John Dewey (1859–1952) proposed that education should be experiential and guide learners to develop

constructive engagement with their surroundings including their larger societal context. Jean Piaget (1896–1980) modeled human cognitive development as proceeding through stages, from basic sensory impressions and motor activities, to a pre-operational stage that includes language acquisition, to concrete operations which include a capacity for logic, and finally to a formal-operational stage marked by abstract reasoning. Lawrence Kohlberg (1927–1987) showed that learners developed moral reasoning in a sequence of stages from self-interest (pre-conventional morality), to social consensus (conventional morality), to universal principles (post-conventional morality). Erik Erikson (1902–1994) conceptualized the life cycle to include a series of thematic tasks whose successive completion was required for subsequent stages of development.

Broadly, considerations for the educator as steward of development are threefold. Probably the most important consideration, the one that has historically been appreciated as maternal wisdom but which is now receiving extensive empirical validation, is that early life exposure to adversity or toxic stress levels confers a major risk for negative outcomes in childhood and beyond, due to disruptive impacts on systems for learning, health, and behavior (Shonkoff et al., 2012). Deleterious effects of repeated biochemical stress reactions have been referred to as allostatic load (McEwen, 1998; Juster et al., 2010), and the effects of childhood adversity on adult outcomes are independent of established adult-status risk factors (Danese and McEwen, 2012). Secondly and from a pedagogical perspective, the educator as steward of development should be sensitive to exposing learners to challenges prematurely. For example, attempts to impart abstract concepts too soon may yield poor grasp of those concepts, while robbing the learner of opportunities for experiences salient to their stage. Thirdly, incomplete maturation or failure to complete a task at a given stage (often in association with adversity or toxic stress) may hinder subsequent progress, just as a weak foundation is problematic for constructing higher floors of a building.

On the basis of scientific consensus and in alignment with the paradigm of allostasis, the importance of a developmental perspective is supported by the Research Domain Criteria (RDoC) initiative of the US National Institutes of Mental Health (NIMH). Decades of research have elucidated relationships between brain functionality and behavior, and the NIMH introduced the RDoC in 2009 as a way to advance understanding of mental and behavioral health by transitioning from symptom and behavior-based diagnostic systems to a framework that focuses on underlying brain biology (Cuthbert and Insel, 2013). The initial core domains of brain functionality identified by RDoC are arousal (including sleep), positive valence (including appetitive or reward-oriented behaviors), negative valence (including fear or anxiety), cognitive systems (for attention, memory, language), and systems for social processes (including affiliation and social communication). These core domains are operative in both health and disease, and they cut across different diagnostic categories. RDoC is poised to support advance in mental and behavioral health by suffusing the approach of researchers and practitioners with conceptualizations and interventions – even those that are entirely psychosocial in

character – that are based on appreciations of brain functionality. Critically, developmental trajectories exist for all the major brain domains defined by the RDoC (Casey et al., 2014). These trajectories are impacted by use and environmental influences, and they can be presumed to influence one another.

Like behavioral health research and care, the field of education may also be poised to benefit from conceptualizing the learning child as a unique being with a unique brain, with core domains in various stages of development. As alluded to in Homeostasis, Allostasis, and Education, the human brain undergoes massive changes from embryogenesis to the post-natal period, and it remains plastic throughout the lifespan, presenting an array of conjecturable implications for educators as stewards of development. Neural systems for executive function and cognitive control do not fully mature until the third decade (Hsu et al., 2014), lending credence to organic educational views that use metaphors of blooming or pruning through late adolescence and beyond. Adolescence is characterized by a marked (quadratic) increase in incentive motivation (Luciana, 2013) that bears upon the developing reward system with implications that have been studied for mental health, but this trajectory might also be explicitly considered and leveraged by educators. Though the existence of a critical period for second language acquisition has been questioned (Vanhove, 2013), early education for bilingualism may confer a variety of benefits for cognitive development, including increased cognitive control and possibly generation of cognitive reserve (Bialystok et al., 2012). Alteration of circadian rhythm or sleep, an aspect of arousal, is common in adolescence and may disrupt executive functionality and reward processing (Hasler et al., 2012; Telzer et al., 2013), and advancing strategy and policy to support optimal sleep across the life span is a sensible priority for any approach to neuro-education (Sigman et al., 2014).

To further illustrate the contrasting approaches of cognitive acquisition-focused education and the neurodevelopmental perspective of the GANE, we consider the hypothetical case of a child who is found to be consistently inattentive, distractible, and fidgety. In a homeostatic, cognitive acquisition learning context, this child may be recommended or referred, by an educator, a physician, or parents themselves, to undergo consultation with a behavioral health specialist. If the child falls outside consensus criteria for normal attention and behavior including executive functionality, he or she may be given a diagnosis of attention deficit-hyperactivity disorder (ADHD). Non-normality may be attributed to a variety of disturbances in underlying neural mechanisms that, though possibly understood to have phenotypic expression along a continuum, nonetheless result in categorical assignment to a status of disease versus non-disease. This evaluation has taken place in a *spatial* dimension, wherein fundamentally the individual is compared, at a given moment in time, to other individuals.

The GANE instead evaluates the child's behaviors or tendencies primarily as a snapshot within a longitudinal frame of complex neurodevelopmental trajectories. That is to say, the GANE is critically sensitive to the *temporal* dimension in its evaluation, such that the individual is appreciated primarily with respect to his or her own past and potential futures. Differences

in the stepwise unfolding of brain domain functionalities may have consequences across trajectories. The pioneering work of Harry Harlow, for example, showed the importance of healthy attachment for later life development as a whole, and a recent report (Roskam et al., 2014) has shown an association between duration of exposure to early attachment deprivation (likely impacting brain domains for social affiliation, arousal, and affective valence) and degree of ADHD symptoms in adolescence. These data reinforce that brain domains interact with one another over time, and that ADHD cannot be conceptualized exclusively as a disorder of executive functioning. Furthermore while individuals with attention deficits may have altered sensitivity in the reward system that may confer a later risk for addictive disorders (Blum et al., 2008), it is possible that this same difference may, for some, contribute to success in risk-related endeavors that can have higher-order societal impact, including entrepreneurship. The GANE developmental view requires a willingness to use the biological imagination, for educators to create mental pictures related to the learner's neural past and future. Such an approach promotes greater respect for the learner's full life, both backward and forward in time, and appreciation of both risks and opportunities.

Implications of a neurodevelopmental perspective are even more significant with respect to interventional strategies. The spatial (cross-sectional) context of homeostatic understanding invokes the need for interventions that can correct putatively dysfunctional neural mechanisms, bringing them in alignment with the average or putative norm. In contrast the temporal (longitudinal) context of the GANE encourages intervention for self-calibrated advance that respects the unique needs of an individual's own given state. In the first place, a nuanced view of the temporal dimension should presume the existence of natural variability for development across all domains of brain functioning, no different from variability in ages for children to undergo physical growth spurts. Such a view should lend a degree of conservatism against interventionist strategies that may have non-trivial risk profiles. More to the point, the GANE educator is at liberty to experiment with ways to support or remediate the learner's attention and behavior – through adjusting educational activities, demand levels, situational contexts, or other strategies – to discover approaches that are effective for the learner's unique needs. Developmental principles may even require regressive movement (“one step back, two steps forward”) that enables re-engagement with earlier tasks or stages that were uncompleted or otherwise disturbed in their unfolding. Such constructive trial and error or complex remediation may be difficult in many state-administered mass educational settings – where requirement for a statically high attention level and expectations for learning are highly standardized and normed – but they are realistic in organic educational traditions, for example homeschooling, the one-room schoolhouse, or other educational settings in which teachers progress with learners from year to year.

The GANE Objective is to Enable the Experience of Authentic Freedom

What makes *Homo sapiens* most distinctive and unique in the animal kingdom is our prefrontal cortex, the brain region that

supports advanced cognitive skills and executive functionality (Goldberg, 2001). The prefrontal cortex mediates capacities for imagination, reasoning, inhibition of impulses, and evaluation of the salience of competing stimuli (both internal and external). The capacity for conscious choice and decision-making permits a qualitatively and quantitatively advanced degree of power over other biological sub-systems, behavior, and the environment at large. As a species we have substantially removed many of the natural pressures which historically influenced our likelihood for survival. Our species status now depends more on our creative interaction with each other and the products of our own prior creation, and our degree of freedom is such that we are now able to influence our own evolution. The prefrontal cortex, mediating the capacity to bootstrap, to re-appraise situations in continuously novel and adaptive ways, to respond rather than react, and to do all the above in the service (if we choose) of higher-order goals, is undoubtedly a critical neural substrate for allostasis itself.

Despite this potentiality, education as homeostatic cognitive acquisition tends to focus on a relatively circumscribed understanding of healthy executive functionality. Modern educational systems typically intend for learners to acquire relatively standardized cognitive skills and knowledge. The GANE instead conceives that executive functionality of a caliber reflective of advanced human creativity and flourishing – as expressed in the ideals of the constructivist – entails a capacity for clarity of perception and thought, discernment, use of the imagination, and exercise of self-regulation and willful action that collectively are likely to require a symphony of functioning across brain domains.

In this context, we highlight the contribution of Rudolf Steiner as a philosopher (which is the basis of his contribution as a constructivist educator). Steiner maintained that a capacity for *intuitive thinking*, in which *concepts* are united with *percepts* (which include feelings), is the prerequisite for *moral imagination*, which is in turn the basis for *ethical individualism* (Steiner, 1894), which he considered to be the highest form of human attainment. For Steiner, the significance of ethical individualism lay in its expression of the human as a truly free being. Education for cognitive acquisition, being society-centric, is least obviously intended as a direct support for an individual's authentic freedom. However, neither can learners limited to organic (or romantic) educational practice be considered truly free, in that they may be bound to a variety of emotions or physical impulses. Even many constructivist thinkers do not imply or adumbrate the advanced conception of freedom articulated by Steiner, in that they may restrict understanding of freedom to political dimensions, or place excess emphasis on social interactional processes or universalistic principles (including Kant's categorical imperative) that may not be consistent with unique and changing exigencies of an individual's particular life and natural context. With its focus on the human power for intuitive thinking – or cognition of concepts in concert with subtle somatosensory perception – Steiner's philosophy of freedom exemplifies expression of competent, integrated, and expansive executive functionality whose emergence is the objective of the GANE.

The form of executive functionality envisioned by the GANE is thus of a variety more nuanced or complex than is commonly presumed adequate or even desirable by cognitive acquisitional education or homeostatic neuroscience, which are substantially aligned with positivist philosophical traditions that discount or reject introspection or intuition as a reliable source for understanding. Cognitive acquisitional education and homeostatic neuroscience tend to view the emotions or intuitions as the province of systematic bias, leading to cognitive error. Or these forms of subjectivity become a matter for consideration only after they have become grossly dysregulated, at which point pathways of clinical mental health evaluation and treatment may be deemed needful. In contrast, the GANE aims to integrate the emotions and interoceptions especially in their subtlety, and to leverage their positive and constructive role for the brain and the learner as a whole. In this regard, meaningful insights appear to be accumulating in the field of affective neuroscience. For example, Antonio Damasio (1994) proposed a “somatic marker” hypothesis which implies that emotional or somatosensory signals are needful for effective abstract reasoning. Interoceptive sensations of one's body state (“gut feelings”) may serve as barometer for one's arousal status, representing signals that have implications for safety or action, and that may be a core element of consciousness itself (Craig, 2009). Furthermore, emotional expression represents critical currency for interpersonal interactions, especially insofar as human relationships are often defined by qualitatively differentiated and calibrated degrees of autonomic arousal (Porges, 2011), and this topic is discussed further in the following section. Sterling (2014) has recently proposed that mental health should itself be defined as “responsiveness of the conscious and unconscious mind to the full range of signals from many sources.”

The objective of the GANE is thus to facilitate the emergence of an advanced form of executive functionality that recognizes, models and supports healthy forms of emotional or intuitive self-awareness, self-regulation and social relationship, and imagination. Less than full optionality to benefit from all these signals, without being overwhelmed by them, represents a deficit of human freedom. Organic as well as constructivist forms of educational practice, with their emphasis on well-being and pro-active interaction, may be well positioned to serve the GANE objective. Cognitive acquisitional educational practices too can support the GANE objective, especially if they include pedagogical strategies that aim for learners to have a *feel* for their acquired skills and knowledge. Normative inclusion of a healthy feeling dimension to educational practice represents a qualitative advance that should have non-trivial consequences for learners' lifelong capacity to interact successfully with their environments. At a minimum, the GANE stands against stressful educational environments that generate an excessively competitive culture or are permissive to social bullying. Such settings will be unlikely to support capacities for subtle somatosensory perception and finely calibrated emotional awareness and expression, and in some cases they may have an outright poisonous influence on learning. This idea is further developed in the following section.

The GANE Strategy is to Proceed with Sensitivity to Rhythms

The insights of the paradigm of allostasis can be restated to the following effect. The brain is the organ that oversees management of the variability of rhythms across systems, and it manages rhythms to increase the overall likelihood of successful interaction with complex environments. Dynamic adaptation or recalibration of system set points for optimal activity patterns is essentially indistinguishable from being in the right rhythm, at the right time, for the right purpose. Rhythmic dynamics exist across different domains of brain functionality, and at different scales of activity within those domains including gene expression, neuronal activation and synaptic neurotransmission, synchronous fluctuation of neuronal assemblies, bidirectional brain–body communication, and subjective experiences and observable behaviors. In particular, we propose that attunement to rhythms of *arousal* is likely to hold key value for the educator, and for purposes of the GANE we discuss arousal as the pattern of activity in the autonomic nervous system.

Historically, the sympathetic (“fight or flight”) and parasympathetic (“rest and digest”) divisions of the autonomic nervous system have been considered to act in paired antagonism. That is to say, these divisions have been conceived as functioning in a manner wherein an individual is either in a state of low or high arousal (parasympathetic or sympathetic activation, respectively). Of the two divisions, the sympathetic has been most commonly associated with states of disturbance, especially with respect to mental health (Roth et al., 2008), pain (Martinez-Martinez et al., 2014), and cardiovascular disease (Seravalle et al., 2014), and increased sympathetic activity has been demonstrated in young women with school burnout (May et al., 2015).

Alternatively, the polyvagal theory models autonomic functionality in a way that advances beyond the notion of paired antagonism between the sympathetic and parasympathetic divisions. To begin, polyvagal theory recognizes the existence of two anatomically and functionally distinct sub-systems of the parasympathetic division that derive from different phases of vertebrate evolutionary phylogeny (Porges, 2011). A myelinated branch of the vagus nerve (the main nerve of the parasympathetic division) is an evolutionarily advanced component of the autonomic nervous system, especially operative in humans, and it functions as a fine-tuned brake on high-arousal (fight/flight) mechanisms of the sympathetic division. The myelinated vagus permits self-calming and nuanced forms and degrees of emotional communication (and thus likely supports the advanced executive functionality that is the objective of the GANE). In contrast, an unmyelinated branch of the vagus, especially operative in early (reptilian) vertebrates, produces a “freeze state” for situations of novelty but also overwhelming stress. The unmyelinated vagus may be associated with neurogenic bradycardia (brain-directed slowing or even stopping of the heart), emotional numbing, or behavioral shutdown.

Polyvagal theory proposes that the functionality of these divisions is organized in a hierarchical way with a basis in evolutionary phylogeny. Normatively and in a non-threatening

environment, the myelinated vagus should prevent wasteful high arousal mediated by the sympathetic division. If an environment is perceived to be dangerous, then myelinated vagal activity may give way to sympathetic fight/flight functionality to permit mobilization behaviors, so that a stressor can be physically overcome or escaped. If sympathetic fight/flight is inadequate, then unmyelinated vagal activity produces a freeze or shutdown mode of last resort. Humans will tend to “neurocept” different degrees of safety in different social and environmental contexts, and they may tend to calibrate their autonomic arousal toward one of these modes of functionality, without conscious intent. For safe, threatening (but tractable), or overwhelmingly stressful environments, autonomic regulation will tend to calibrate, respectively, toward parasympathetic fine-tuning, fight/flight arousal, or a parasympathetic freeze mode. All three modes or levels of arousal recognized by polyvagal theory are conserved in humans, and the different modes have pervasive influence on numerous aspects of behavior, performance, and health that are salient to the GANE.

In the first place and as a matter of primary pedagogical strategy, allostatic neuro-education aims for experientially palpable and subjectively enjoyable oscillation between parasympathetic fine-tuning and subtle degrees of sympathetic arousal. We propose that a rhythm of educational practice that is sensitive to alternation in these autonomic states has greater potential to educe the learner’s engagement, and in self-directed ways. Periods of excitement may ebb and flow with periods of calm reflection or incubation, in association with interoceptive functionality of the vagus nerve as described by polyvagal theory, to support awareness of one’s visceral sensations. Moreover, polyvagal theory recognizes that the brainstem nucleus of the myelinated vagus overlaps with cranial nerves that manage muscles for facial and vocal emotional expression, and auditory sensation. Descending activation of the myelinated vagus may support enhanced capacity for highly nuanced emotional communication with others. Thus parasympathetic fine-tuning, associated with safe environments, enables calm and subtle bidirectional brain–body communication and should facilitate appraisal of information in undelimited ways, to increase the learner’s cognitive engagement and optionality of response. It seems likely that a range of influences on executive functionality including physical movement (Khalil et al., 2013; Rommel et al., 2013), musical training (Kraus et al., 2014; Miendlarzewska and Trost, 2014; Fonseca-Mora et al., 2015; Francois et al., 2015), complex games (Kim et al., 2014), and exposure to natural environments (Taylor and Kuo, 2009) entail rhythmic or playful engagement with the learner’s autonomic arousal level, and these studies further highlight how brain functional domains interact in complex ways to produce lived and meaningful experience.

Secondly, polyvagal theory posits a need to recognize that not all low arousal is created equal. There is a major qualitative difference between the low arousal of an individual who has developed robust executive functionality for fine-tuned parasympathetic (myelinated vagal) self-calming in the face of stress, for example, and the evolutionarily ancient parasympathetic (unmyelinated vagal) freeze mode of someone who has not recovered from trauma. The former individual has

leadership potential, whereas the latter is an individual who is likely to have low emotional self-awareness and may be impaired in their capacity for healthy social interaction. Calibration of autonomic activity for unmyelinated vagal freeze mode may be a pernicious if unrecognized reality in mass educational systems. This mode is likely to be problematic for learning, and it may be associated with emotional disengagement to the degree that it creates risk for maladaptive compensatory behaviors including anti-sociality (Lee et al., 2014).

The GANE strategy of attunement to rhythms of learning may have implications for reconsidering the burgeoning use of computing technologies in education. Computers have been widely promoted in education on the generally presumptive basis that the scope and speed of information afforded should translate into benefits for educational practice. In contrast, others voice caution about the use of technology in education, and this view is expressed in Steiner-inspired education. Steiner schools explicitly refrain from exposing learners to computing technology until the later middle grades or high school, on grounds that such influence may produce risk for detrimental effects on the learner as a whole. Instead the learning environment is focused on GANE themes introduced earlier, with attention to the child as a composite of multiple domains, using rhythm-sensitive exercises, creative arts and crafts, foreign language, music, natural environments, and physical movement, in ways that make maximal benefit of direct human engagement, in alignment with the implications of polyvagal theory, and with sensitivity to developmental context. Like Steiner schools, the GANE emphasizes the value of real-time human-to-human engagement as a way for the learner to use the teacher as a mirror or a guide to develop their capacity for autonomic fine-tuning and stress responsivity.

Nonetheless, neither the poly-vagal perspective nor the GANE imply that the computer *per se* is a negative influence in education. The key questions, as with all technology, instead pertain to whether and how computing technology can be best put to the most thoughtful and constructive use. For example, we hypothesize the likelihood of educational value from orchestration of real-time interactions between young learners in a rural environment and geographically distant teachers of a foreign language. Such computer-enabled learning interactions might add invaluable sophistication to education as cognitive acquisition (or cultural transmission), and also could support the GANE to achieve its highest constructivist objectives.

Implications of the GANE

Testable Hypotheses Related to the GANE

The GANE is not itself a theory in the sense of being a unitary and falsifiable conjecture. Rather it is a re-visioning of what education might represent, and how it might more faithfully serve both learners and society. The perspective that favors the learner's long-term, whole-brain, whole-life trajectory, is a value choice. Likewise the objective of expansive executive functionality, aligned with Steiner's contention that the highest human good that can be attained is ethical individualism, is a philosophical premise. Many learners, educators, and parents

who gravitate toward the GANE may do so on the basis of their resonance with its principles, rather than because of empirical validation in controlled studies.

Nonetheless a variety of meaningful hypotheses relating to the core elements of the GANE can be generated and tested. Its strategy to attend to *rhythms*, and especially rhythms of arousal and experiential excitement, is perhaps the most amenable to relatively straightforward scientific exploration. The familiarity of this idea suggests it is a general-purpose strategy that could serve a variety of educational approaches as well as vocational needs. Numerous studies, including those mentioned in Section "The GANE Objective is to Enable the Experience of Authentic Freedom," suggest that thoughtful inclusion of activities involving rhythmic behavioral or sensory attunement – for example physical education and athletics, music, drama, dance, and related pursuits – as elements of core curriculum, may enhance rather than detract from the achievement of educational objectives including those related to cognitive acquisition. Moreover, an expansive conception of "rhythm" invites hypotheses that explore highly technical understandings of *brain* rhythms and how they may relate to learning, and this topic is explored in Section "Case Studies Illustrating Use of Allostatic Neurotechnology."

A range of hypotheses may be inspired by the long-term developmental perspective of the GANE. Studies may focus on developmental causes of a learning challenge, rather than symptomatic remediation. In particular, the role of toxic stress described in Section "The GANE Perspective is the Learner's Extended Neurodevelopmental Trajectory" would appear to be a critical area for further investigation. It has been proposed that early childhood adversity and toxic stress should be approached with the seriousness that is accorded to major medical risk factors, and that pediatric medicine should take a leadership role on this front (Shonkoff et al., 2012). Sensitivity to the influence of stress on development should, however, also be an obvious priority in educational settings, where children spend far more time. Educators are well-positioned to lead or collaborate on hypothesis-driven studies intended to prevent or mitigate the consequences of toxic stress for the brain, body, learning, and behavior.

It is also possible to design studies that aim to identify the respective consequences of educational approaches that place varying degrees of emphasis on long-term developmental trajectories. For example in an evaluation of children enrolled in schools with different reading instruction ages (age 7 in a Steiner-based school, versus age 5 in a state-curriculum school), it was reported that by age 11 there were no reading fluency disadvantages for the children who began reading instruction at the later age (Suggate et al., 2013). Hypothesis-driven studies may also evaluate whether the presence of certain characteristics that are considered non-normative at a given time, may be associated with other forms of positive outcome at a later time in the learner's trajectory.

The GANE objective of enabling the experience of authentic freedom is fundamentally philosophical, nonetheless it also entails testable hypotheses. For example, it has been hypothesized that learners educated on the basis of Steiner principles (or

congruous educational systems) would have more self-efficacy than those learning on the basis of conventional (cognitive acquisitional) curricula, when transitioning to higher education (Shankland et al., 2009). The GANE objective also aligns with increasingly sophisticated hypotheses and experimental paradigms at the intersection of cognitive, affective, and social neuroscience, that aim for more detailed understanding of the multi-directional influences among bodily interoceptions, social interactions or cultural context, and cognitive processing and appraisal (e.g., Immordino-Yang et al., 2014). GANE educators who have a robust qualitative appreciation for learners in their multi-dimensionality (physical, emotional, mental, spiritual aspects) may be well positioned to dialog with neuroscientists to help formulate testable hypotheses that are thoughtfully sensitive to these domains. They may also be positioned to help develop study designs intended to identify brain states that educators might then use to help guide their pedagogy (Gabrieli et al., 2015).

Case Studies Illustrating Use of Allostatic Neurotechnology

We contend that technological innovation can ease the path of allostatic neuro-education if the technology is itself guided by allostatic principles. Allostatic technology should aim to support individuals to be optimally adapted to their environmental context, in part by supporting system set points to be dynamically flexible and not fixed. Given the complexity of the brain, its dynamics, and its role for central command, allostatic technology aims to respect the brain's own capacity for self-optimization, which may be expected to involve increased efficiency for learning. More technically, it has been proposed that allostatic interventions should restore responsiveness of neural systems to the full range of signals from many sources, and to do so by using "natural mechanisms for predictive regulation [that] involve continual updating of knowledge stores" (Sterling, 2014). By contrast, brain-centric technologies exist including electroencephalographic biofeedback ("neurofeedback"), transcranial magnetic stimulation, transcranial direct current stimulation, and others that are largely homeostatic in their reliance on normative (and externally given) standards for neural functioning, and/or strategy based on direct induction of corrections or changes to brain mechanisms. In this section we present two case studies that illustrate use of a non-invasive allostatic neurotechnology in ways that are aligned with the GANE and may support the formulation of controlled, hypothesis-driven studies that relate to the GANE and supportive technologies.

Case 1

An 18 years-old male, carrying diagnoses of Asperger's syndrome and ADHD, was enrolled in an IRB-approved, open label exploratory study at Wake Forest School of Medicine, evaluating the use of an allostatic neurotechnology for a range of different purposes. The technology operates through rapidly updating the brain about its own oscillatory activity through the medium of sound and is intended to support brain activity to be flexibly adaptable around an individual's unique oscillatory set points (Gerdes et al., 2013). The participant's mother reported that

he was always, "on edge, hyper-focused, and a light sleeper." He attended several local private schools for elementary and middle school years, including one focused on students with learning challenges. He had been home-schooled for the last several years, and had just completed the school year at the time of enrollment. He had been using the stimulant lisdexamfetamine dimesylate (Vyvanse®), 30 mg daily, for management of attention deficit symptoms, for 2 years prior to enrollment. Under his physician's guidance the stimulant was tapered to 1–2 times per week, and was then discontinued just prior to study entry. Supplements included astaxanthin, omega 3, blue-green algae, calcium, magnesium, and Co-Q 10.

After informed consent, the participant completed self-report symptom inventories including the Insomnia Severity Index (ISI), Beck Anxiety Index (BDI), Beck Depression Index – II (BDI-II), and the Autism Spectrum Quotient (AQ). An assessment of brain electrical frequencies and amplitudes was obtained, as previously described (Gerdes et al., 2013), consisting of 3 min recordings obtained from standard locations on the scalp (based on the 10–20 system), and including 1 min of recording for each of three eye states (eyes closed, partially closed, and eyes open). With eyes closed, the participant was at rest, while a cognitive task was performed during eyes open recording. The assessment provided a "snapshot" of relative symmetry between homologous brain regions, along with the distribution of amplitudes among different frequency bands at each location.

Scores on baseline symptom inventories – ISI 6, BAI 14, BDI-II 11, and AQ 39 – suggested absence of clinically relevant insomnia, presence of mild anxiety, presence of depressive symptoms on the upper end of the normative range, and a strong likelihood of Autism Spectrum Disorder. Result for the BDI-II also included an elevated score for the measure assessing Concentration Difficulty ("It's very hard to keep my mind on anything for very long"). **Figure 2** shows baseline assessment spectrographs of brain electrical activity based on 1 min recording at the frontal pole locations (FP1, left; FP2, right) with eyes open. The very low frequency ranges (0–1 Hz) were notable for amplitudes of 9.8 and 10.3 μV at FP1 and FP2, respectively. At the temporal locations (T3 left, T4 right) with eyes closed (**Figure 3**), baseline assessment suggested 39% T4 dominance in the high frequencies (23–36 Hz), with average amplitudes of 1.7 and 2.4 μV on the left and right, respectively, in the same frequencies.

The participant undertook 14 sessions with the allostatic neurotechnology (13 days of sessions, median of 76 min, range 64–84, with total protocol time of 1,066 min) over a total period of 40 days, with a 27 days recess between sessions 10 and 11. Sessions were well tolerated, with no adverse events reported. **Figures 4** and **5** provide spectrographs of data from the penultimate minute, of the penultimate session, providing insight into shifts that occurred during the course of the intervention. Electrical amplitudes were reduced across the frequency spectrum at bilateral frontal poles, and in the very low frequency (0–1 Hz) range they were an average of 2.8 μV bilaterally. At bilateral temporal lobes there were reduced amplitudes in the 23–36 Hz range (0.36 μV on left, 0.32 μV

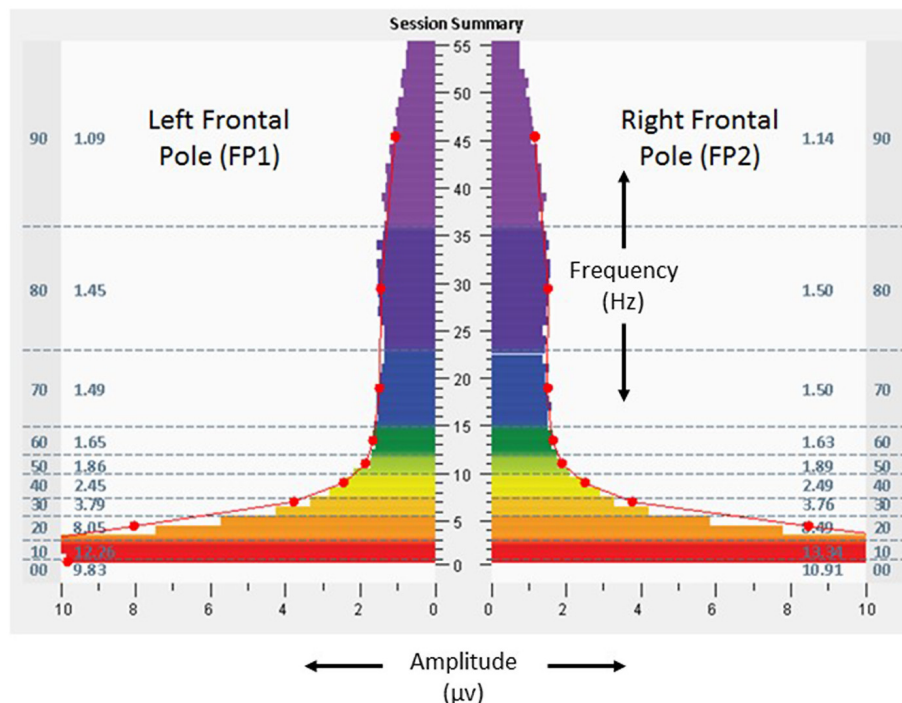


FIGURE 2 | Fast Fourier Transform spectral display of 1 min recording of brain electrical activity at left and right frontal poles (FP1 left, FP2 right), with eyes open, during the baseline assessment, from the 18-years-old learner described in the text. Individual color bars reflect amplitude averages for 1 min of recording, eyes closed, at rest, without

stimulation. Columns to the left and right of the color bars denote 10 frequency ranges of aggregated data (00: < 1.0 Hz; 10: 1.0–3.0 Hz; 20: 3.0–5.5 Hz; 30: 5.5–7.5 Hz; 40: 7.5–10.0 Hz; 50: 10.0–12.0 Hz; 60: 12.0–15.0 Hz; 70: 15.0–23.0 Hz; 80: 23.0–36.0 Hz; 90: 36.0–48.0 Hz) and numerical values for amplitude averages in those ranges.

on right), with 13.2% left dominance at the same frequencies, representing a reduction in asymmetry.

At the post-intervention follow-up data collection visit (12 days after completion of sessions), the participant offered that he “felt better” and that “something seemed different.” His mother reported, “His teachers for summer programs noted his concentration and focus were better in class, as well as his willingness to participate.” The mother also noted that he was sleeping better and more soundly. He remained off the stimulant medication. Follow-up scores on symptom inventories – ISI 1, BAI 13, BDI-II 1, and AQ 40 – reflected improvement in sleep and reduction of depressive symptoms. For the BDI-II measure on Concentration Difficulty, the participant no longer endorsed concentration as a problem. Additional telephone follow-up with the mother, 4.5 months after completing the intervention, revealed that the participant was now enrolled in a special education program at a local high school, which included mainstream classes with the general school population. He had joined the local chapter of the ROTC and was being praised for his performance. She reported that the participant had remained off of the stimulant for 3 months, but that he had been re-started at 15 mg per day, half the prior dose, 5 days per week, as school started.

As discussed in Section “Perspective, Objective, and Strategy of the GANE,” homeostatic neuropsychiatry views attention-deficit hyperactivity disorder as a categorical diagnosis

reflecting aberrant neural mechanisms. Alternatively, the GANE views cognition and behavior as being expressions that pertain to an individual within the context of their unique neurodevelopmental trajectory. The adolescent in this case was supported with approaches that reflect both strategies – educational settings aimed for sensitivity to the role of environmental factors on learning, and also use of stimulant medication aimed to increase (clamp) attention toward a more normative set point. In this case study, adjunctive use of allostatic neurotechnology was associated with self-adjustments in cognitive systems (attention), arousal (sleep), and social affiliation (class participation). High amplitudes in a very low frequency range (0.02–0.2 Hz) of neural oscillations have been described as an endophenotype of ADHD that relate to the default mode network (Broyd et al., 2011), and stimulant use in adults with ADHD has been associated with reduction in inattention and attenuation of very low frequency amplitudes (Cooper et al., 2014). Neural oscillatory changes represented as reductions in low frequency amplitudes at bilateral frontal poles in this individual may have been related to improved capacity for attention. Increased amplitudes in high frequency ranges have been described as a basis for insomnia (Perlis et al., 2001), raising the possibility that improved sleep quality in this subject was related to reductions of amplitude in the high frequency (23–36 Hz or higher) ranges demonstrated in both frontal pole and temporal regions. The participant’s baseline rightward

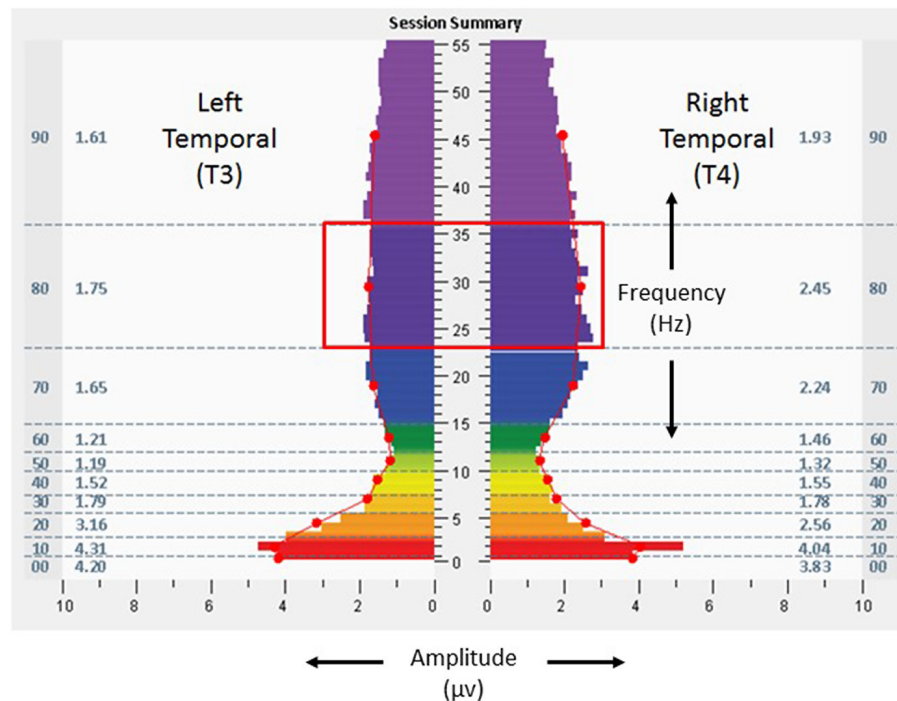


FIGURE 3 | Fast Fourier Transform spectral display of 1 min recording of brain electrical activity at the left and right temporal lobe (T3 left, T4 right), with eyes closed, during the baseline assessment, from the 18-years-old learner described in text. Red box denotes the 23–36 Hz frequency range mentioned in the text. See **Figure 2** legend for detailed explanation of data elements.

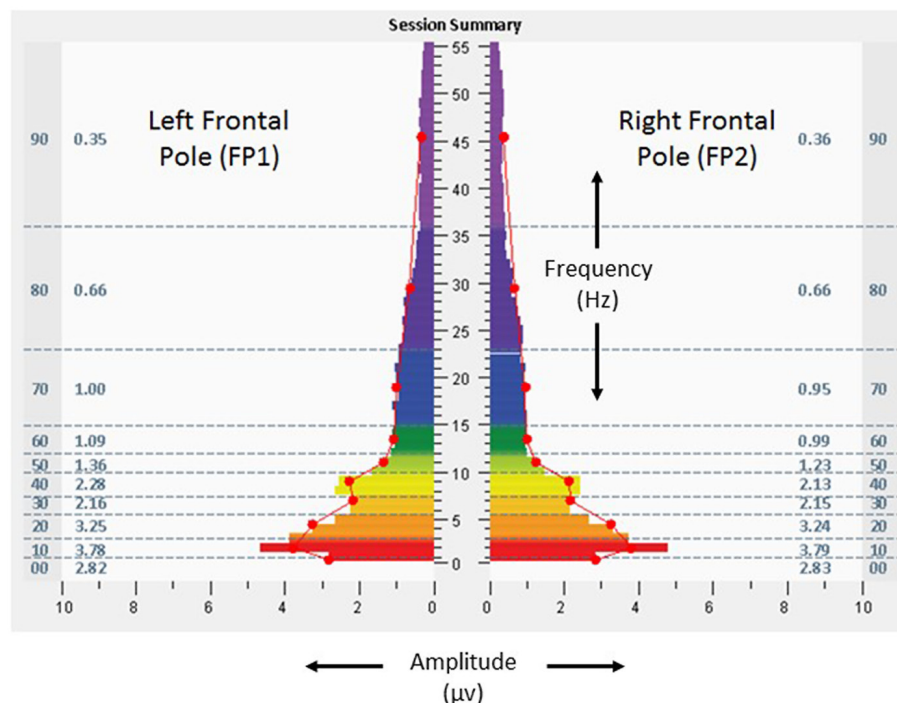


FIGURE 4 | Fast Fourier Transform spectral display of brain electrical activity observed during the penultimate minute of the penultimate allostatic technology session at the left and right frontal poles (FP1 left, FP2 right), with eyes open, from the 18 years-old learner described in the text. See **Figure 2** legend for detailed explanation of data elements.

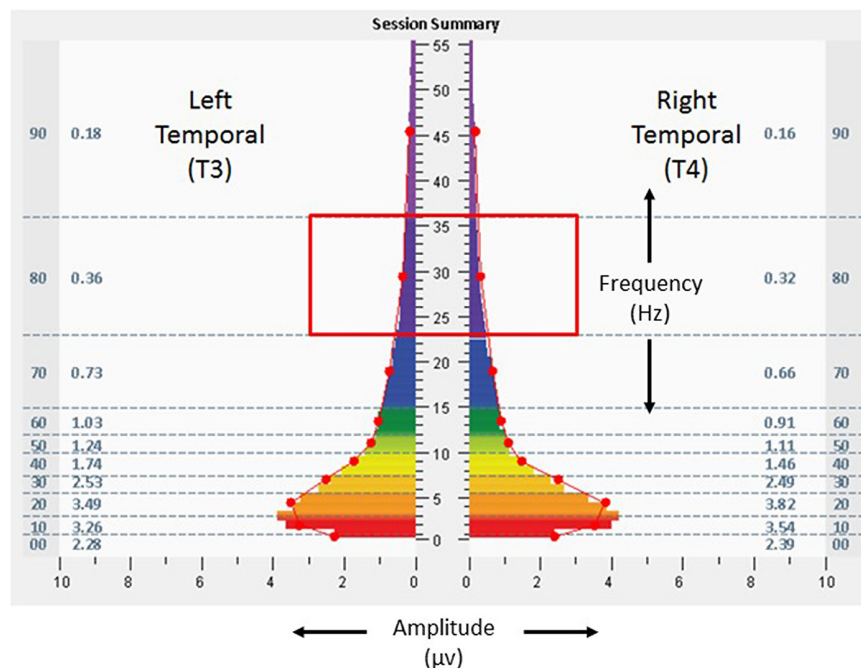


FIGURE 5 | Fast Fourier Transform spectral display of brain electrical activity observed during the penultimate minute of the penultimate allostatic technology session at the left and right temporal lobe (T3 left,

T4 right), with eyes closed, from the 18 years-old learner described in the text. Red box denotes the 23–36 Hz frequency range mentioned in the text. See **Figure 2** legend for detailed explanation of data elements.

dominance in temporal high frequency electrical asymmetry may have been suggestive of relative sympathetic tendency in autonomic regulation (Tegeler et al., 2015), and reduction of this asymmetry may have reflected calibration of his stress responsivity toward greater parasympathetic (myelinated vagal) self-regulation.

Stimulant medications have effects on neurodevelopmental trajectories for mood, stress responsivity, and reward processing (Marco et al., 2011; O'Daly et al., 2014). They are neurotoxic (Goncalves et al., 2014) and appear to impair neuroplasticity (Urban and Gao, 2014), and parents are shown to have apprehensions about their use for ADHD (Ahmed et al., 2013). In contrast to this cautionary view, others have proposed that homeostatic neuro-pharmacological intervention may be salutary for development, on the basis that targeted drug intervention may reprogram developmental trajectories so as to enable preventative cure of conditions such as ADHD and depression (Andersen and Navalta, 2011). To date, long-term follow-up of children treated for ADHD has not shown benefits for titrated psychoactive stimulant usage (or behavioral intervention, or their combination) over community-based care, on either academic, social, or clinical mental health outcomes (Molina et al., 2009).

Case 2

A 9 years-old girl in her fourth grade at a public school was noted to have “inadequate response to individualized educational interventions” and so was referred for evaluation by an educational diagnostician. She was assessed to have a

specific learning disability related to reading and was placed in a Special Education Program. Her mother was advised that she should expect for her daughter to remain in this program for the duration of her formal schooling (through high school). The following summer she experienced a traumatic emotional loss when her maternal aunt died, at which time according to her mother she began experiencing emotional difficulties, including tendencies for anger and being “closed off.” Her medical and behavioral health history included enuresis (bed-wetting) and seasonal allergies. She used no medications. In the spring of her fifth grade, the child's mother arranged for a series of 14 sessions using the same allostatic neurotechnology described in Case 1 (Gerdes et al., 2013), in the hope that doing so would support improvement in her reading comprehension, and also for possible emotional benefits. Initial effects noted by the mother included a greater degree of calm, being “not so wound up,” and more openness. Within about 9 months after the initial sessions, the mother felt that her learning capacity had increased markedly, with improvements demonstrable as faster speed for processing information. Behaviorally, the mother noted that the child had no episodes of enuresis for 9–10 months after starting sessions, followed by a recurrence of episodes that appeared to be related to anticipation of the end of school. The child underwent five more sessions in the winter of sixth grade. By the end of seventh grade, the mother's impression was that the child was demonstrating significant improvements in reading comprehension. She was composing reports easily and in ways that included advanced word uses, and could communicate orally without stuttering. In the beginning of

her eighth grade, the child's skills were evaluated to be in a range that did not require the Special Education Program, and she began the year in a regular eighth grade classroom. Her initial marks (August 28, 2014) were English B+, Mathematics B, Physical Education A+, Science B+, and Bilingualism A+.

The child in Case 2 appeared to demonstrate a combination of behavioral and learning difficulties that may have been exacerbated by an emotional trauma, highlighting the influence of life events across brain domains, and possibly indicating that she was stalled at a developmental stage. Adults for example with complicated grief have been found to have deficits in cognitive function and structural brain changes (Saavedra Pérez et al., 2015). Nocturnal enuresis is reported to be associated with autonomic dysregulation (Yakinci et al., 1997). In this case study, use of allostatic neurotechnology was associated with improvement in emotional well-being as well as relief from enuresis, and these shifts are conceivably related to recovery from effects of emotional stress (Lee et al., 2014) that may have had implications for learning ability.

Summary and Conclusion

Early and other approaches to neuro-education have used the metaphor of a bridge (Bruer, 1997; Sigman et al., 2014), which implies a fixed distance between the educator and the neuroscientist. In contrast, the present paper proposes that educational leaders and brain specialists who are like-minded in a developmentally sensitive and constructivist orientation, can collaborate now on groundwork that supports a new vision of learning. The GANE adduces the evolutionary model of physiological regulation known as allostasis, to flexibly apply a range of educational practices including the organic (home-schooling and other child-centric methods), cognitive acquisition (attainment of common understandings, measured through testing), and the constructivist (to enable dynamic forms of individual-societal interaction and renewal). Allostatic neuro-education recognizes change and not constancy to be the norm, defines success within the context of complex and changing natural environments as opposed to controlled settings, and identifies the brain as the organ of central command. In this paper we have characterized the GANE by introducing its perspective, its objective, and its strategy. The perspective is to focus on the learner's full neurodevelopmental trajectory, rather than the immediate dictates of standardized testing. The objective is to support the emergence of competent, integrated,

and expansive executive functionality, that supports the highest expression of humans as free beings. The strategy is to guide learning with attention to rhythms, especially rhythms of arousal, and to do so in ways that support palpable excitement. A variety of testable hypotheses derive from the GANE, and there may be particular need for studies of ways to prevent or mitigate the consequences of childhood adversity or toxic stress. Allostatic neurotechnology may support the GANE by respecting the brain's complexity and supporting its capacity for self-optimization.

Life at any time is unpredictable, and much of its wonder is that the most unpredictable and unimaginable events are the ones most likely to produce the changes of genuine significance (Taleb, 2007). Recognition of this reality may lend greater weight to the constructivist aphorism that "the best way to predict the future is to create it." Serious appreciation of unpredictability furthermore compels us to prepare new generations of learners – and their developing brains – to develop capacity to be adaptable in new ways and to greater degrees. In contrast, promotion of education as a way to support economic growth may be dubious strategy. Not only does such emphasis tend to demoralize both learners and educators, empirical study has found that, contrary to much public policy discourse, increasing education does not lead to increased economic productivity (Pritchett, 2001). In context of the accelerating change currently being experienced across societal sectors and around the globe, the aim of the GANE is to breathe new life into educational practice by educating the full potential of learners, through constructive appreciation of the complex and ever-changing human brain.

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Review of neural rehabilitation programs for dyslexia: how can an allophonic system be changed into a phonemic one?

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Neural investigations suggest that there are three possible core deficits in dyslexia: phonemic, grapho-phonemic, and graphemic. These investigations also suggest that the phonemic deficit resides in a different mode of speech perception which is based on allophonic (subphonemic) units rather than phonemic units. Here we review the results of remediation methods that tap into each of these core deficits, and examine how the methods that tap into the phonemic deficit might contribute to the remediation of allophonic perception. Remediation of grapho-phonemic deficiencies with a new computerized phonics training program (GraphoGame) might be able to surpass the limits of classical phonics training programs, particularly with regard to reading fluency. Remediation of visuo-graphemic deficiencies through exposure to enhanced letter spacing is also promising, although children with dyslexia continued to read more slowly than typical readers after this type of training. Remediation of phonemic deficiencies in dyslexia with programs based solely on phonemic awareness has a limited impact on reading. This might be due to the persistence of a covert deficit in phonemic perception. Methods based on slowed speech enhance the perception not only of phonemic features but also of allophonic features, and this is probably why they have not been found to be effective in meta-analyses. Training of phonemic perception with a perceptual fading paradigm, a method that improves precision in identification and discrimination around phonemic boundaries, has yielded promising results. However, studies with children at risk for dyslexia and dyslexic adults have found that even when behavioral data do not reflect allophonic perception, it can nevertheless be present in neural recordings. Further investigations should seek to confirm that the perceptual fading paradigm is beneficial for reading, and that it renders perception truly phonemic.

Keywords: dyslexia, determinants of dyslexia, allophonic perception, rehabilitation, perceptual fading

THE THREE SOURCES OF DYSLLEXIA

Developmental dyslexia is a specific learning disability characterized by difficulties in the acquisition of low-level reading skills: i.e., accurate and/or fluent word recognition and decoding skills (Lyon et al., 2003). Developmental dyslexia affects about 5–10% of the population (Shaywitz et al., 1990; Peterson and Pennington, 2012). Low-level reading skills, especially decoding skills, are chiefly a matter of relating the basic units of the written language (letters and groups of letters called graphemes) to the basic units of the spoken language (phonemes). Children with dyslexia experience great difficulties in learning grapheme–phoneme associations and, once acquired, these associations remain suboptimal (Lachmann and van Leeuwen, 2014). Dyslexic children do not read fluently and expend much more energy in reading than typical children (Shaywitz and Shaywitz, 2005; Sprenger-Charolles et al., 2006; Blomert and Vaessen, 2009).

The most obvious possible reason for dyslexics' problem in establishing grapheme–phoneme relationships is a deficiency in cross-modal neural mechanisms (Blomert, 2011). Evidence of areas responsive to the simultaneous presentation of letters and speech sounds in the temporal cortex (superior temporal sulcus, STS and superior temporal gyrus, STG) has been presented. Furthermore, it has been shown that when a letter and sound occur within the same narrow time-window, letters influence the processing of speech sounds (van Atteveldt et al., 2004). This and other related findings (Blomert and Froyen, 2010) suggest that letter–sound integration is performed by specialized neural processes. Such cross-modal integration also occurs in dyslexic children with 4 years of reading instruction, but the influence of print on sound perception is much weaker for them than for age-matched controls, and it only appears when the letter is presented much earlier than the sound (Froyen et al., 2011).

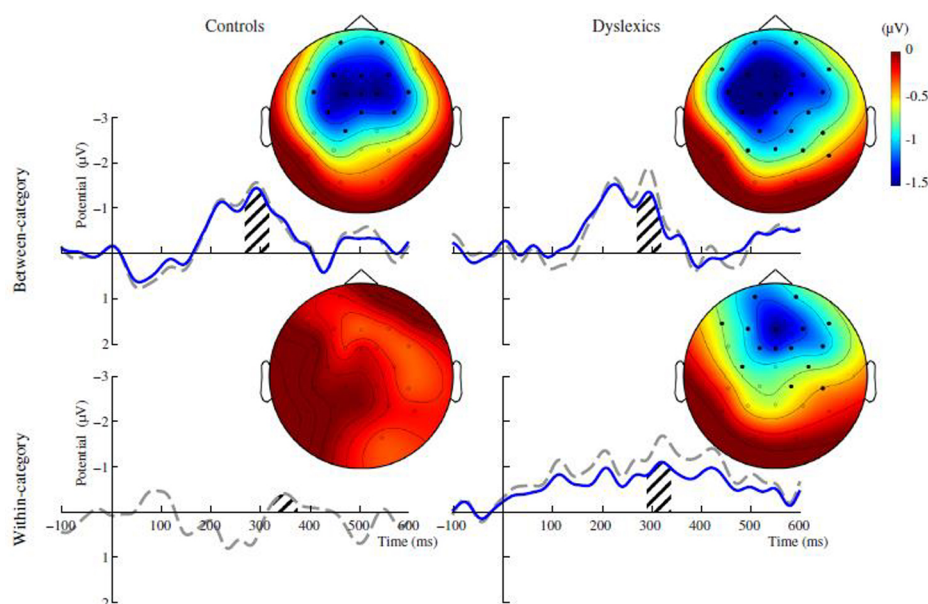


FIGURE 1 | Mismatch negativity (MMN; a pre-attentive neurophysiological response, a component of the event-related potential) for a phonemic contrast (above) and for an allophonic contrast (below) in adult dyslexics (right) and controls (left). Adapted from Noordenbos et al. (2013, Figure 3).

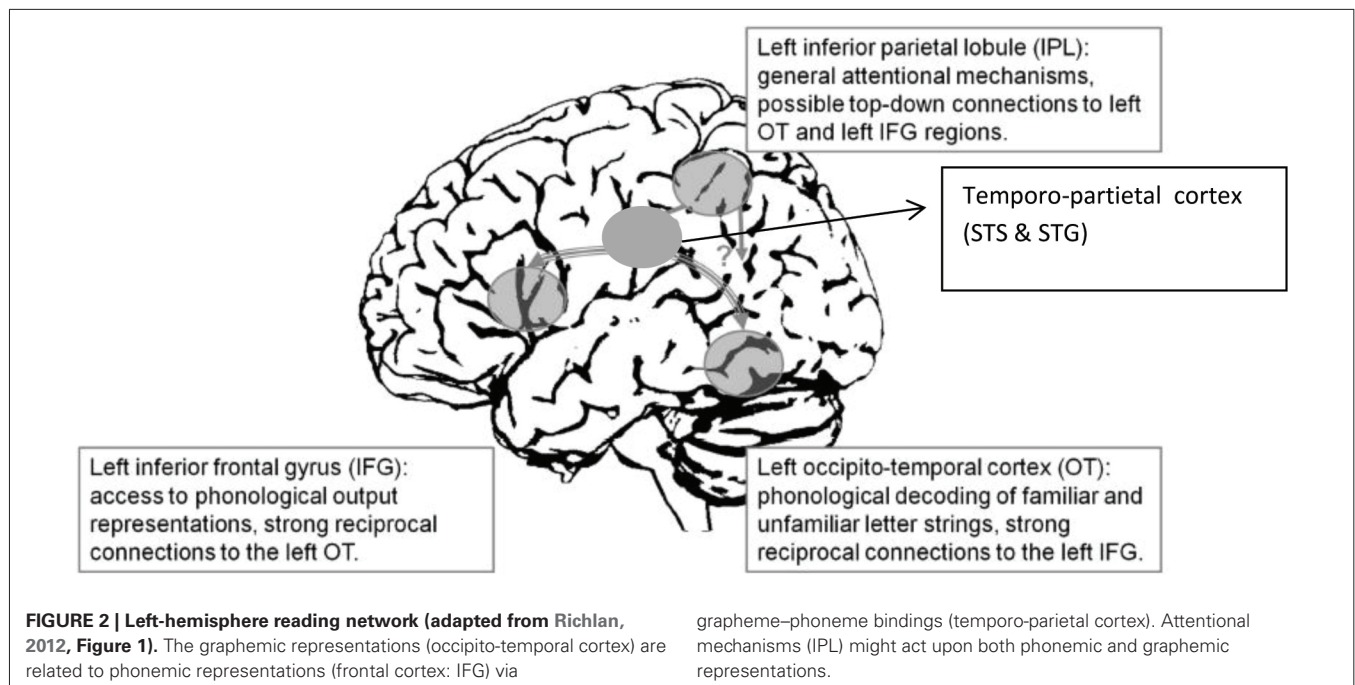
A specific failure in the simultaneous binding of letters with speech sounds is not the only possible cause of dyslexia, however. There are two other main reasons why grapheme–phoneme associations might be deficient in the absence of specific binding problems. The first factor that might also affect grapheme–phoneme associations is a deficiency in the visual processing of letters¹. This hypothesis has been formulated in various different ways, and might be explained in the framework of a recent theory (the “neuronal recycling hypothesis”: Cohen and Dehaene, 2004; Dehaene, 2014). The theory attributes fluent word recognition to a specific brain area of the left hemisphere [dubbed the “visual word form area (VWFA)”], which was initially devoted to visual processing requiring a level of acuity similar to that needed by letter processing but which, in recent human history, has been recycled for letter perception. However, a recent survey indicates that, besides being used for visual word perception, the VWFA has maintained its original function in processing other visual stimuli (Vogel et al., 2014; see also Dehaene, 2014). Vogel et al. (2014) also noted evidence that activity in the occipito-temporal cortex is strongly correlated with the dorsal attentional network. This is in accordance with several studies that point to the role of visuo-attentional deficits in part of the dyslexic population (e.g., Franceschini et al., 2012; Lobier et al., 2012).

A second factor that might also affect grapheme–phoneme associations is a deficiency in the phonological processing of

speech sounds. Children with dyslexia often exhibit a lack of phonemic awareness: i.e., a problem with the ability to segment words into phonemes, a skill which is required to learn to read in an alphabetic system, but not required to learn to speak (Liberman et al., 1974). A deficit in phonemic awareness might be responsible for difficulties in relating these units to graphemes (for a review, Melby-Lervåg et al., 2012). However, the deficit in phonemic awareness is probably the consequence of a more drastic difference in the mode of speech perception. Perceiving speech sounds in terms of subphonemic units (allophones) induces serious problems for relating them to phoneme-sized graphical units. This is the possibility raised by the “allophonic” theory of dyslexia (Serniclaes et al., 2004).

There is growing evidence that individuals with dyslexia discriminate between allophonic variants of the same phoneme, whereas typical-reading controls do not perceive such distinctions (Bogliotti et al., 2008; Noordenbos et al., 2012a,b, 2013, for a comprehensive review of the available evidence; Serniclaes and Sprenger-Charolles, 2015). Even when there is apparently no behavioral manifestation of allophonic discrimination (e.g., Messaoud-Galusi et al., 2011), it can nevertheless be present in the brain. This has been evidenced by the results of studies conducted in Dutch with either children at risk for dyslexia or adults with dyslexia (Noordenbos et al., 2012b, 2013; see **Figure 1**). The lack of a behavioral manifestation of allophonic processing, that in fact takes place at the neural level, suggests the involvement of inhibitory processes. Such processes would inhibit the neural responses to allophonic contrasts so that only the neural responses to phonemic contrasts would be available for emitting the behavioral responses. According to a PET study with French adults with dyslexia, such processes might take place in the frontal cortex in the inferior frontal gyrus (IFG) close to

¹One of the possible indicators of a visual source of dyslexia is the absence of problems in non-word reading, which is fairly rare in the dyslexic population (see Sprenger-Charolles et al., 2011, for a review of the literature, with a meta-analysis of 300 cases of dyslexics). However, the phonological deficit might be a secondary consequence of a more basic visual deficit, and there are also positive indicators of visual problems, such as those in parallel letter-string processing (Lobier et al., 2012).



Broca's area (Dufor et al., 2009). Inhibition is costly in terms of metabolic resources that then are not available for reading, a possible cause for the slow and laborious performance in word recognition and decoding that characterize dyslexia (Shaywitz and Shaywitz, 2005; Sprenger-Charolles et al., 2011). Reduced metabolic resources on reading might for instance slower the transmission of the phoneme percept from the frontal cortex to the areas of the temporo-parietal cortex that are responsible for grapheme–phoneme associations.

In summary, the processes involved in low-level reading skills are carried out by a neural network (**Figure 2**) that relates graphemic representations (occipito-temporal cortex) to phonemic representations (frontal cortex) via grapheme–phoneme bindings (temporo-parietal cortex). In turn, the three sources of dyslexia could be summarized as follows: a grapho-phonemic deficit due to a lack of strong and timely grapheme–phoneme associations, a graphemic deficit due to a failure to combine letters (or graphemes) into word representations, and an audio-phonemic deficit arising from an allophonic mode of speech perception.

Each of these three possible core deficits has prompted attempts at remediation. Here we first review the results of the available remediation methods. We then see how the remediation of allophonic perception might contribute to overcome some of the limitations of these methods.

REMEDICATION OF GRAPHEME–PHONEME ASSOCIATIONS

As a failure to associate graphemes with phonemes is the most proximal cause of dyslexia, intervention studies should primarily aim to enable or improve the learning of grapho-phonemic associations. Not surprisingly, then, different kinds of grapho-phonemic training have been used in attempts to aid in reading acquisition and remediate dyslexia.

The results of a first meta-analysis (Ehri et al., 2001a, see also the results of the long-term longitudinal study of Johnson et al., 2012) indicate that systematic phonics instruction (mainly when based on grapheme–phoneme correspondences and not on rhyme units for instance) can improve the acquisition of low- and high-level reading skills, especially when training begins early and in children at risk for reading disability; the benefits of such training are lesser in children with reading disabilities (dyslexics). However, the results of two recent meta-analyses of training studies (McArthur et al., 2012; Galuschka et al., 2014) indicate that classical phonics instruction is the only treatment approach whose efficacy in children and adolescents with reading disabilities is statistically confirmed. Furthermore, as noted by Gabrieli (2009), only about 50% of dyslexics retain the reading progress they make after explicit and systematic instruction in decoding strategies and phonemic awareness, and those who do retain their gains do not attain the fluent reading competency of typical-reading children.

A new computerized phonics training program (Grapho-Game) appears to be able to surpass the limits of classical phonics training programs, especially with regard to reading fluency. The aim of GraphoGame is to strengthen the binding between the orthographic and phonological encodings of words (for a review, Richardson and Lyytinen, 2014). The GraphoGame method is mainly based on the training of grapheme–phoneme correspondences. This method progresses from the simultaneous and repeated presentation of grapheme–phoneme correspondences (first in isolation, then included in syllables, and afterward in words) to fluency training with words and sentences. GraphoGame's effectiveness in improving reading acquisition has been demonstrated in several studies conducted in languages with various levels of orthographic transparency: Finnish

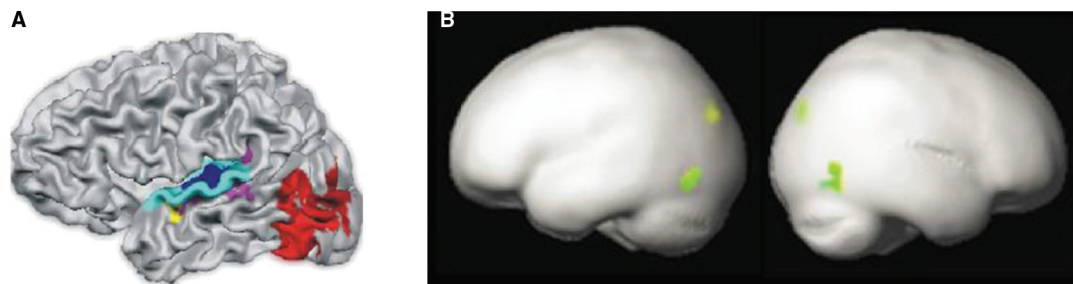


FIGURE 3 | (A) Letter-sound association area (in blue) and VWFA (in red) in the left STS-STG (adapted from Blomert, 2011, Figure 1). **(B)** GraphoGame training effects (in green; adapted from Brem et al., 2010, Figure 2).

(Saine et al., 2010, 2011), German (Brem et al., 2010), and English (Kyle et al., 2013).

In the Finnish study, after a screening of 166 first graders [with tests assessing letter knowledge, phonological awareness, and rapid automatic naming (RAN) of letters, digits, or pictures of frequent words], the lowest-achieving 30% were randomly assigned to two different remedial interventions (25 children in each group): a regular remedial phonics intervention (RRI; Note²) or a computerized assisted intervention using GraphoGame (computer assisted regular remedial intervention, CARRI), both with four weekly sessions of 45 min for 28 weeks. These two groups were compared to the remaining children, who received “mainstream” reading instruction. For word reading fluency, at the end of the first grade, the CARRI group outperformed the RRI group, while both differed from the mainstreamers. One year later the difference between the CARRI group and the RRI group was still significant, but not the one between the CARRI group and the mainstreamers. Moreover, at that time, only three children from the CARRI group (11%) still presented a severe deficit in word reading fluency, versus 11 children from the RRI group (44%). Although these data are drawn from on a small number of participants, they are of interest, especially those from the RRI group, which are very similar to those reported by Gabrieli (2009) about the percentage of dyslexics who are “resistant” to classical phonological interventions. In addition, the CARRI group’s gradual gains in word reading fluency indicate that children at risk for reading disability can reach the level of mainstream students. However, they require much more time to reach that level. These Finnish results were replicated in a language with a non-transparent orthography (English, Kyle et al., 2013). However, the interpretation of the results of that study is limited by the fact that no individual data were provided and no fluency evaluations were performed.

In another training study (Brem et al., 2010), functional magnetic resonance imaging (fMRI) data were collected from 16 German-speaking kindergarteners from Switzerland trained with both GraphoGame and a non-linguistic number-knowledge control game (duration of each of the two training programs: less than 4 h per week over 8 weeks). The results showed that

behavioral improvements were accompanied by activity changes in the VWFA in the left occipito-temporal cortex (Figure 3). This contrasts with (later) findings by Blomert (2011) showing that the neural site of letter–speech sound bindings is located in the left temporal cortex suggesting that the results of a grapho-phonemic training method such as GraphoGame should primarily affect the letter–sound area. An effect of grapho-phonemic training on the VWFA is not surprising because the development of that areas depends on reading instruction (Dehaene et al., 2010). However, it nevertheless seems that GraphoGame should primarily impact the neural site of letter–speech sound bindings. One possible explanation to this discrepancy is that in Brem’s study the effects of GraphoGame were assessed by comparing sensitivity to letters vs. other visual symbols. Possible changes in the sensitivity to letter–sound bindings, that should take place in temporo-parietal cortex, were thus not directly evaluated.

Instead of directly arising from a deficit with complex written symbols, the deficit in visual-auditory integration might arise from remote lower-level deficits. Training 7-year-old dyslexic children to associate elementary sound features (e.g., duration) with simple visual features (e.g., length) has been found to have positive effects on reading skills (Kujala et al., 2001). A recent study evidenced impaired audio-visual integration of low-level stimuli in dyslexic adults (Harrar et al., 2014). However, another recent study with adolescent dyslexics found that they exhibited specific problems with grapho-phonemic conversions even though their basic audio-visual integration mechanisms seemed to be intact (Kronschabel et al., 2014). Whatever conclusions are ultimately drawn on this point, remediation methods might benefit from a better understanding of the processes involved in grapheme–phoneme integration.

REMEDIATION OF VISUO-GRAPHEMIC DEFICIENCIES

There have been several attempts to remediate graphemic deficits in dyslexia through the facilitation of low-level visual processing. A study with Italian and French children with dyslexia (Zorzi et al., 2012) showed that simple exposure to enhanced letter spacing led to improved reading accuracy and speed in both linguistic groups (34 to 40 children with dyslexia per language group, about 10 years old, 2-month follow-up). However, a subsequent study with Spanish children (Perea et al., 2012) found that the reading speed of children with dyslexia after exposure to enhanced letter

²The RRI included activities linking reading, spelling and phonology, training of word segmentation, training of decoding and spelling, and vocabulary training.

spacing remained lower than that of typical readers (18 children with dyslexia, about 12 years old). This limitation might be due to the fact that the existing visuo-graphemic interventions do not tap into the perception of letters as visual categories. Letter enhancement taps into low-level visual processing, so that it magnifies not only the distinctions between letters that are relevant for word decoding, but also a host of graphical details that do not contribute to letter recognition.

Other remediation studies have aimed at improving visuo-attentional performance using video games (e.g., Green and Bavelier, 2012). Franceschini et al. (2013), used “action” and “non-action” video games, differing in cognitive load and speed requirements, and compared their effects on reading. Two groups of 10 Italian children with dyslexia, of about 10 years of age, were randomly assigned to the “action” and “non-action” training groups (12 h at 80 min per day). The results showed significant improvement in reading performance only with the “action” training. When measured with a speed/accuracy score, the resulting progress in reading was equivalent to one year of spontaneous reading development. However, the study participants did not seem to present phonological deficits, meaning that the benefits of visuo-attentional training cannot be generalized to the whole dyslexic population. More importantly, studies in this area are still too rare to allow generalizations.

REMEDICATION OF PHONOLOGICAL DEFICIENCIES

INTERVENTIONS AIMED AT IMPROVING PHONEMIC AWARENESS

Meta-analyses indicate that early phonemic awareness training helps children at risk for reading disability to acquire word-level reading skills, but such training has lesser effects in those who have already developed reading difficulties (Ehri et al., 2001b; National Institute for Literacy, 2008). These meta-analyses also highlight the fact that such training is very effective only when the letters (or graphemes) are presented together with the corresponding phonemes: remediation methods that train phonemic awareness alone have a limited impact on reading, and especially fluent reading. Furthermore, interventions using both grapheme-phoneme training and phonemic awareness training have neural effects in the left hemisphere reading network (Démonet et al., 2004), including the VWFA (Brem et al., 2010).

INTERVENTIONS AIMED AT IMPROVING LOW-LEVEL AUDITORY PROCESSES

Numerous auditory training methods have been proposed (for a review, Collet et al., 2014). For instance, Earobics® (Morrison, 1998; Diehl, 1999) is a computer-assisted training program which aims to improve reading skills by improving children's sound perception, memory, and phonological awareness. This program consists of a number of tasks, such as phoneme identification and discrimination and rhyme judgments. It has been widely used in the teaching of reading in American schools, but also in children with language learning difficulties specifically. Using this program with dyslexic children, Russo et al. (2005) showed a significant improvement of neural synchrony in the auditory brainstem in children who had received the training, while those who had not received this training showed no such changes. These results suggest that dyslexic children derive some benefits from

this training, and that these benefits are also seen at the level of subcortical structures.

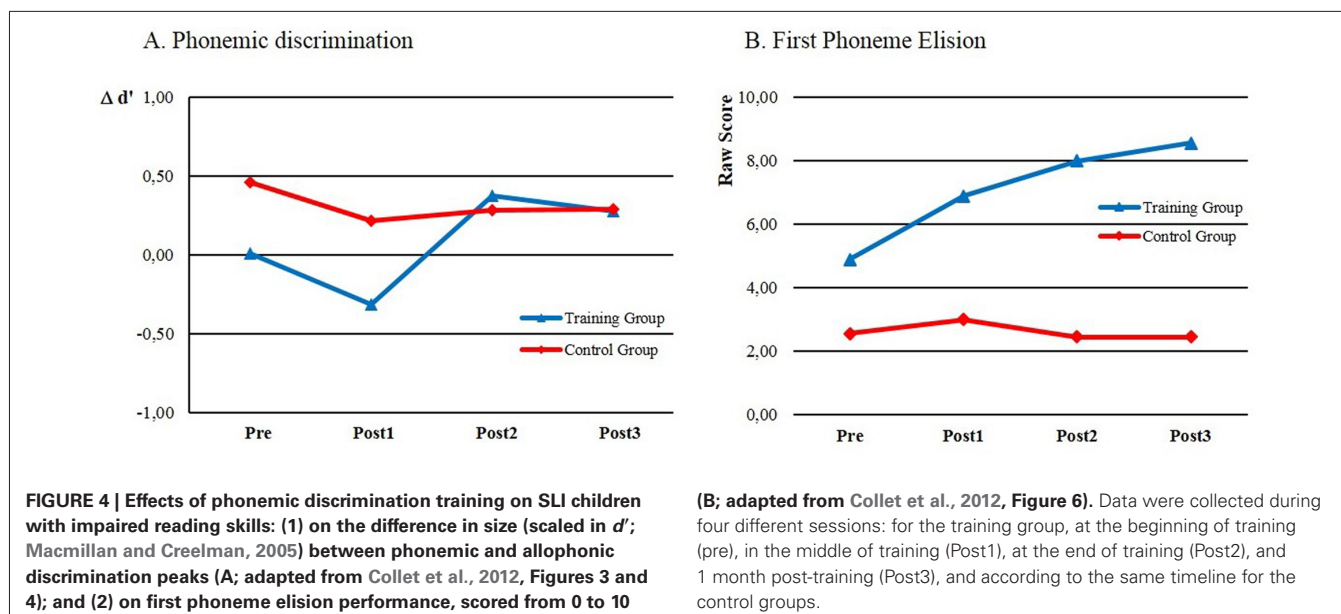
In the same vein, other studies have attempted to develop procedures to improve the auditory-perceptual abilities of children with learning disabilities. Merzenich et al. (1996) and Tallal et al. (1996), hypothesized that dyslexic children have a temporal processing disorder that could be remediated through auditory training, developed a computerized training program known as Fast ForWord® (Scientific Learning Corporation, Oakland, CA, USA). The program consisted in a succession of tasks such as the comparison or identification of sounds, phonemes, syllables, and words with variations in acoustic parameters such as duration and frequency, and was recommended during a period of 6 to 8 weeks (100 min a day, 5 days a week). Tallal et al. (1996) found that such auditory training had positive effects on the perception and understanding of speech. However, meta-analytic reviews indicate that these remediation attempts do not have reliable effects on reading performance (e.g., Strong et al., 2011).

INTERVENTIONS AIMED AT REMEDIATING ALLOPHONIC PERCEPTION

Contrary to typical phonemic perception, which combines different auditory features and weights them differently as a function of contextual features, allophonic perception uses these features independently and irrespective of context. For instance, French dyslexic children are sensitive to two different features that are perceived independently by the pre-linguistic child but are dynamically combined for separating voiced and voiceless consonants in French, with weights depending on the syllabic context (Bogliotti et al., 2008). Remediation of allophonic perception is intrinsically difficult, because it means modifying processes that allow the child to perceive speech sounds, albeit in a non-optimal way. There is no need to tap into auditory processes to remediate allophonic perception, because the use of allophonic units is not a matter of auditory feature perception as such but a matter of combining auditory features in way that is relevant for speech perception.

Discriminant training of minimal pairs (Hurford, 1990; Hurford and Sanders, 1990; Veuillet et al., 2007) might be of some help, but it has no straightforward implications for phonemic perception. Discriminating two different phonemes is supposed to be achieved with a phonemic boundary, but it can also be achieved through one of the several allophonic boundaries that separate these two phonemes. Similarly, the deletion of the initial phoneme from a word and other “phonemic awareness” performances are normally achieved with a phonemic cut-off point, but they can also be achieved with allophonic cut-off points. What is needed to remediate allophonic perception is to modify the boundaries that are used to discriminate and segment speech sounds, something that is not guaranteed with classical methods.

Until now, only a handful of studies have tried to remediate allophonic perception in people affected by dyslexia. Bogliotti (2005) trained severely impaired dyslexic children (five trained children and five untrained controls between 8 and 10 years of age) to identify allophonic variants of the same phoneme with the same label (following a procedure initiated by Guenther et al., 1999). The training improved the accuracy of phoneme identification, but it did not improve discrimination around the



phoneme boundary. On the contrary, the training gave rise to discrimination peaks around allophonic boundaries. Allophonic discrimination was probably present in these children before training, but it only became apparent in behavioral responses after training. This suggests that allophonic perception is indeed highly resistant to training.

Recently, Collet et al. (2012) developed a new method, adapted from the “perceptual fading” training program (Jamieson and Morosan, 1986). The basic approach was to progressively reduce the acoustic distance between two stimuli as a function of each child’s individual performance. This method aimed to teach children to discriminate fine acoustic differences between two different phonemes. During the study, the stimuli varied along a $d\partial/t\partial$ voice onset time (VOT) continuum, and the acoustic difference in VOT around the French VOT boundary was progressively reduced. At each stage, these pairs of different phonemes were mixed with other random pairs composed of identical phonemes. The task required the child to determine whether the pairs sounded alike or different. After each answer, the child received positive or negative visual feedback (green or red screen) on accuracy. As soon as the child’s performance was stabilized above 75%, the acoustic distance between phonemes was reduced in the next training step. This transition thus occurred when minimally distinct phonemes in acoustic terms were discriminated above chance level.

The total duration of the training was about 18 h (2×9 sessions of about 25 min each). Eighteen 9-year-old children with specific language impairment (SLI; which delays the mastery of oral language skills) who also had impaired reading and spelling skills (at least 1.5 standard deviations below the mean for their age) participated in the study. These children were randomly assigned to either a training group or a control group of equal size. Results showed that perceptual fading improved both discrimination and identification performance in these children. Allophonic discrimination peaks emerged after the initial training sessions, just as in the previous study with dyslexic

children (Bogliotti, 2005, see above), but they were progressively replaced by phonemic peaks in the later sessions (Figure 4). Importantly, phonemic awareness considerably improved after perceptual training. Unfortunately, reading performance was not evaluated at the end of the training.

Several important questions remain open concerning the impact of phonological remediation with perceptual fading (hereafter audio-phonological remediation, APR). One question is whether APR contributes to remediating reading deficits in dyslexia. The fact that the SLI children studied by Collet et al. (2012) also began with a reading deficit suggests that APR will also improve phonemic awareness in dyslexic children. And although there is presently no (published) evidence in support of the benefits of APR for reading, given the strong effects of APR on phonemic awareness it should also have at least some impact on reading performance. Preliminary results from APR training with dyslexic children suggest that this type of training is indeed beneficial for reading and spelling performance (work in progress).

Another question is whether APR truly transforms an allophonic system into a phonemic one. Recall that discrimination of allophonic peaks can be completely absent from behavioral responses even when it is present in neural processing (in children: Noordenbos et al., 2012b; in adults: Noordenbos et al., 2013). APR might thus give rise to a hybrid system that appears to be phonemic but that remains basically allophonic. Still another question is whether APR is beneficial for individuals with dyslexia who do not exhibit allophonic perception at the behavioral level although their neural processing is allophonic. Studies examining neural activity are needed to clarify these points.

CONCLUSION

Among the various methods that have been used in attempts to remediate dyslexia, those involving grapho-phonemic training are currently the most successful. However, as there are three possible sources of dyslexia (phonological, grapho-phonemic, and

graphemic) several different methods need to be tried. Graphemic methods are successful in part of the dyslexic population. Phonological remediation based on phoneme awareness alone has only a limited effect on reading, especially in dyslexic children. A possible reason for these limitations is that training a child to manipulate phoneme-like segments does not guarantee a change in the way the child perceives these segments. Some recent studies suggest that a subset of people with dyslexia perceive speech in allophonic segments instead of phonemic ones, a distinction that is not captured by phoneme awareness tasks. A new method of phonological remediation that is specifically designed to change an allophonic mode of speech perception into a phonemic one is promising, although its effects on reading need to be confirmed.

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Hearing and music in unilateral spatial neglect neuro-rehabilitation

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Unilateral spatial neglect (USN) is an attention deficit in the contralesional side of space which occurs after a cerebral stroke, mainly located in the right hemisphere. USN patients are disabled in all daily activities. USN is an important negative prognostic factor of functional recovery and of socio-professional reinsertion. Thus, patient rehabilitation is a major challenge. As this deficit has been described in many sensory modalities (including hearing), many sensory and poly-sensory rehabilitation methods have been proposed to USN patients. They are mainly based on visual, tactile modalities and on motor abilities. However, these methods appear to be quite task-specific and difficult to transfer to functional activities. Very few studies have focused on the hearing modality and even fewer studies have been conducted in music as a way of improving spatial attention. Therefore, more research on such retraining needs is necessary in order to make reliable conclusions on its efficiency in long-term rehabilitation. Nevertheless, some evidence suggests that music could be a promising tool to enhance spatial attention and to rehabilitate USN patients. In fact, music is a material closely linked to space, involving common anatomical and functional networks. The present paper aims firstly at briefly reviewing the different procedures of sensory retraining proposed in USN, including auditory retraining, and their limits. Secondly, it aims to present the recent scientific evidence that makes music a good candidate for USN patients' neuro-rehabilitation.

Keywords: unilateral spatial neglect, rehabilitation, sensory retraining, hearing, music

INTRODUCTION

Unilateral spatial neglect (USN) is a neuropsychological syndrome characterized by an attention deficit in the contralesional side of space (Posner et al., 1984). USN cannot be linked to a sensory or a motor deficit. USN patients fail to orient themselves toward contralesional targets. Many spatial deficits can appear with USN patients: colliding with left side objects while walking, dressing only one side of their body or failing to eat the food on the neglected side of their plate. Thus, they are severely disabled in all daily activities. This syndrome results mainly from a right hemispheric damage after a stroke (Bartolomeo et al., 2012). Cerebral lesions could be located in a large territory in the brain, going from the parietal lobe to the frontal one. Stone et al. (1991) found that 72% of patients with a right cerebral stroke had USN 3 days after. After 3 months, 33% of these patients still showed signs of neglect signs which tended to last for years. This deficit is an important negative prognostic factor of functional recovery (Held et al., 1975; Denes et al., 1982). Therefore, its rehabilitation is a major challenge as USN could negatively influence motor recovery and also social and professional reintegration.

A variety of clinical tests exist to assess USN. Assessment usually relies on "paper and pencil" tests. The most common conventional tests are the line bisection tasks (Halligan and Marshall, 1991), the cancellation tasks, such as the star cancellation

task (Wilson et al., 1987) and the bells test (Gauthier et al., 1989). Simple drawing, copying of figures, reading and writing tasks are also used. In each of them, the number of reported elements located in the contralesional side of space is considered in order to evaluate the presence of USN. However, these tasks have several limitations. For example, they are essentially based on visual modality and they do not identify the daily life difficulties of patients. Some batteries of tests have been designed in this aim. The Behavioural Inattention Test (BIT; Wilson et al., 1987) is the most commonly used test and consists of both the aforementioned "paper and pencil" tests and behavioral procedures to evaluate USN. However, its ecological validity remains questionable: behavioral procedures are not more sensitive than conventional tests to detect USN (Halligan et al., 1991). Therefore, an evaluation of daily life activities appears to be necessary to evaluate the functional impact of USN. However, only a very few scales exist, such as the Catherine Bergego Scale (Bergego et al., 1995; Azouvi et al., 2003). Due to the lack of assessment tools, this aspect is often forgotten during clinical assessments.

Although USN is essentially evaluated in the visual modality in clinical practice, deficits of spatial attention in USN have been described in many other sensory modalities: touch (De Renzi et al., 1970; Barbieri and De Renzi, 1989), hearing (De Renzi et al., 1989; Pavani et al., 2001), and proprioception (Vallar et al., 1993).

Barbieri and De Renzi (1989) characterized tactile neglect as a failure to detect tactile stimulations on the left side of a patient's body. In hearing, USN patients may show errors in localizing and lateralizing sounds (Pavani et al., 2001) often shifting them toward the right side. In addition, they may present real auditory neglect with a failure to detect a stimulus perceived with the left ear (De Renzi et al., 1989). Vallar et al. (1993) also showed that USN patients have a deficit in their position sense when compared to right damaged patients without USN signs. In other words, USN patients have difficulties in localizing themselves in space. Moreover, in USN, motor neglect with a spontaneous under-use of the contralesional arm could also be observed without hemiplegia or other deficits (Laplane and Degos, 1983; Coulthard et al., 2008).

Even though there are only a limited number of studies on USN in modalities other than the visual modality (particularly because of the lack of clinical tools to assess them), USN seems to affect all sensory modalities and not only vision. De Renzi et al. (1970) suggest that USN is caused by mutilated space representation and this can affect all the sensory modalities. Therefore, USN can be interpreted as a supramodal spatial bias even though some differences of severity between modalities can be found (Chokron et al., 2002).

In the next section, we will conduct a review of the main sensory rehabilitation methods based on the theoretical conception that USN is a spatial attention deficit involving different sensory modalities. In the following sections, after underlying the contribution of hearing in USN rehabilitation, we will present the recent scientific evidence that makes music and musical practice promising tools for USN patients' rehabilitation.

SENSORY RETRAINING

The visual scanning training method (Diller and Weinberg, 1977; Pizzamiglio et al., 1992) is the most commonly used method in clinical practice. The underlying theory of this method is that USN is a spatial exploration deficit and retraining this top-down processing can lead to rehabilitation. Patients learn to voluntarily pay attention to the left side of visual space due to an initial target placed on their left side. This target is used as a visual anchoring point and its salience decreases session after session so that USN patients have to pay more and more attention by themselves. Various tasks such as reading tasks or cancelation tasks can be used with this method. The complexity of these tasks can also be enhanced during training sessions by increasing the number of distractors in the cancelation task or reducing the letter size of the text in the reading task. This rehabilitation provided quite good results on USN neuropsychological tests (Diller and Weinberg, 1977; Pizzamiglio et al., 1992). However, this rehabilitation used similar tasks to those included in the neuropsychological assessment, therefore preventing a generalization of the training effects in everyday life (Seron et al., 1989; Wagenaar et al., 1992). The gains due to this retraining appear to be task-specific and not transferable to functional activities.

Other rehabilitation methods exist and are based on the hypothesis that USN is an impairment of coordinate transformation used to represent extra-personal space and can be

rehabilitated by recalibrating the perception of space. These rehabilitation techniques aim at modifying cognitive maps to induce more accurate behaviors. The cases of optokinetic stimulations or prismatic glasses in the visual modality are examples of such techniques. In other modalities, caloric or transcutaneous electrical stimulations, also rely on this hypothesis. Optokinetic stimulations are based on displays of visual stimuli all moving coherently on a computer screen to the patient's neglected side. Some studies (Vallar et al., 1993; Karnath, 1996) showed improvements in visual scanning in the neglect field and an improvement in the deficit in their position sense with this method. According to Kerkhoff et al. (2006), the presentation of moving visual stimuli with active smooth pursuit eye movement can be more efficient than the conventional training of visual scanning. Finally, in a study by Pizzamiglio et al. (2004), one group of patients received only visual scanning tasks as a form of rehabilitation whereas another group received visual scanning tasks combined with optokinetic stimuli. This latter situation did not show more efficiency than visual scanning using only static stimuli. Moreover, these studies did not evaluate the potential effects on daily life. Prismatic glasses, which modify the perception of visual environment so as to induce a gaze deviation to the left, are another method to rehabilitate USN patients (Rosetti et al., 1998; Serino et al., 2009). These prismatic glasses generally deviate the visual field 10° to the right. Unlike in the visual scanning training method, the prismatic glasses modify perceptual environment in order to change cognitive maps. They use bottom-up processing to rehabilitate USN. In this treatment, patients wear prismatic glasses and have to go through target-pointing tasks. This device has shown promising results. Unfortunately, these results dure often only in the short-term [in the study by Rosetti et al. (1998) the results dured almost 2 h]. Prismatic glasses involve a habituation mechanism to induce a change in the cognitive maps. When the patient removes the glasses, the spatial habituation continues for a certain time. However, the visual perception without glasses will involve a new change in cognitive maps with a return to the previous and pathological ones. This explains the short-term efficiency of this treatment. Furthermore, some studies (Rousseaux et al., 2006) did not find any effect (for a review, see Rode et al., 2006).

Other rehabilitation methods using the vestibular and the somato-sensory modalities have also been used when treating USN. These methods are based on the same hypothesis of the two previous techniques and aim to calibrate cognitive maps. Vestibular stimulation, also known as caloric stimulation, involves the use of cold water in the left ear (Rode et al., 1998). In this method, caloric stimulation has been found to reduce USN symptoms. However, this remission is temporary, only lasting from thirty to sixty minutes. Transcutaneous electrical stimulation can also be used. Vallar et al. (1995) showed that a left neck electrical stimulation could improve performance on line or letter cancelation tests in 13 of the 14 USN patients included in their study. These two previous methods are efficient in treating USN symptoms. However, the major issues are similar to those faced with in visual scanning training methods and concern generalization to daily life and long-term efficiency.

These methods yield important but temporary remission of deficits.

Other approaches target motor abilities in order to help the rehabilitation of visual attention. Using motor rehabilitation (Robertson et al., 1992), it was shown that a contralesional arm mobilization could reduce USN signs for several weeks after treatment. Robertson and North (1992) underlined that a contralesional arm activation in the contralesional hemi-space was more successful in reducing USN signs on a cancellation task than one in the other hemi-space.

Finally, many rehabilitation techniques have been proposed on sensory modalities other than the visual one but difficulties in evaluating the impact of these techniques were reported as we have previously seen. Indeed, the clinical tools used to evaluate the efficiency of a rehabilitation method are essentially based on the visual modality, and overall, do not take into account the possible improvement in other modalities and, rarely, in daily life. In order to come closer to real life improvements, studies using poly-sensory rehabilitation methods have emerged.

POLY-SENSORY RETRAINING

Since we live in a multi-sensory environment, combining modalities bring the rehabilitation closer to real life. Brunila et al. (2002) chose to use motor skills with left arm activation combined with visual scanning training in order to help the latter and showed considerable improvement on cancellation tasks. Nevertheless, there was no improvement found in other tasks. In their design, only one rehabilitation patient group was used. This group participated in a training program which combined left arm activation with visual scanning. The authors concluded that the gain effect could have been caused by an efficient combination of the two methods, visual scanning training and left arm activation. However, the observed effect also could have been caused by the efficiency of only one of the two previous methods. This study could not dissociate the two hypotheses as no control group was used. In order to answer this question, Luukainen-Markkula et al. (2009) compared the visual scanning training method to the left arm motor activation rehabilitation method. They underlined that the efficiency of the two programs were roughly the same and that both could have been useful in the approach of Brunila et al. (2002). These authors used this finding to argue that combine modalities and efficient rehabilitation methods could be a better way to rehabilitate USN.

Recently, Polanowska et al. (2009) combined a left-hand somato-sensory electrical stimulation with the visual scanning training method. Their study demonstrated that patients who received the combination technique had better visual exploration results for than those who received only the visual scanning training method. The authors argued that the electrical stimulations involved an activation of the attention system of the right cerebral hemisphere and would help visual exploration. Therefore, multi-sensory retraining appears necessary. However, there was no ecological test and no functional evaluation, except the Barthel Index (Mahoney and Barthel, 1965). This index is not specific to USN patients but was created to evaluate physical disabilities and did not show any improvement

in this study. Difficulties for daily life generalization can still persist, as the previous methods showed, which are difficult to underline by the tests used in a clinical approach. Developing and using more ecological tests appears necessary to evaluate the daily life use of the poly-sensory retraining program, even though, it provides better results than retraining based on only one sensory modality. Finally, it is important to highlight that no rehabilitation based on more than two sensory modalities was found in the literature and that poly-sensory retraining always involves the visual modality. The improvement of the other modalities after rehabilitation is often under-assessed and these modalities are also less used in USN rehabilitation.

CONTRIBUTION OF HEARING IN RETRAINING

And what about hearing? In literature, very few studies take an interest in the potential effects of auditive stimulations in USN rehabilitation. This is probably due to a lack of audition assessment tools. However, some studies have provided evidence that auditory stimuli could significantly enhance visual perception in USN. These patients show an improvement in visual detection when visual and auditory stimuli come from the same position in space, unlike when they came from two different positions (Frassinetti et al., 2002a,b). This improvement is greater for visual positions affected by the USN, that is to say in the left part of space most of the time. These studies have underlined the existence of an integrated visuo-auditory system in USN patients and the importance of using auditory stimulations in order to help visual detection.

Furthermore, some evidence of auditory stimulations as an effective modality training for USN patients exists. Hommel et al. (1990) proposed that, in USN patients, music stimuli could be superior to other sensory or cognitive cues (such as speech or tactile cues). In their study, listening to binaural non-verbal auditory stimuli decreases USN symptoms. Patients reported more elements in the left side in a copying drawings task, whereas binaural auditory verbal stimuli and unilateral or bilateral tactile stimuli have no effect on this task. This effect was found with classical music such as with white noise, suggesting that this effect is not specific to music. The effect can be linked to a greater activation in the right cerebral hemisphere compared to the left one, whereas it was not the case with verbal stimuli. According to the authors, verbal stimuli imply a bilateral hemispheric activation and a persistence of interhemispheric imbalance in USN patients. These results underlined that auditory cerebral pathways could take part in the network connected to USN and non-verbal passive auditory stimuli could improve USN symptoms. Therefore, this could have a real relevance in regards to circumvent visual modality as deficits could be major in vision and hearing seems to be a valid alternative.

Nevertheless, contrary to all other sensory modalities, no study was really focused on the specific effect of the auditory stimulations in long-term rehabilitation. Some studies used auditory stimulations combined with other sensory rehabilitation methods. Yet, in these rehabilitation methods, auditory stimuli were only used as feed-back information or as alerts but not as a real rehabilitation tool (Fanthome et al., 1995; Robertson et al., 1998)

with an analysis of the specific effects of auditory stimulations on USN symptoms.

MUSIC IN USN REHABILITATION

Music as an auditory stimulation is particularly interesting to examine. This is because, as we see in Hommel et al.'s (1990) study, music activates more the right cerebral hemisphere than the left one and, therefore, could improve neglect.

Recently, studies showed that just listening to music could improve spatial attention in USN (Soto et al., 2009; Chen et al., 2013; Tsai et al., 2013). The visual awareness on the contralesional hemi-space increased when visual tasks were performed with patient's preferred music relative to their un-preferred music or silence (Soto et al., 2009). Preferred music enhanced the detection and the identification of contralesional targets in a perceptual report task, enabled patients to make more accurate midline bisection judgments and improved the reading on the contralesional side of space. However, only three patients were included in this study. Chen et al. (2013) did a similar experiment on nineteen patients and also established that listening to pleasant music has a positive impact on three subtests in the BIT (the Star Cancellation Test, the Line Bisection and the Picture Scanning Test) and, so may improve visual attention in USN patients. Similarly, results in Tsai et al. (2013) indicated that listening to a classical music increases performances on the same three subtests of the BIT. Music appears to be a promising tool to rehabilitate USN.

Even with the possibilities presented, music as auditory stimulations has not yet been used as tool for long-term rehabilitation in USN. From our review of the literature, every few studies using music material to rehabilitate USN patients was found. However, music has more frequently been used in long-term rehabilitation of patients with a cerebral stroke. Sarkamo et al. (2008) conducted a study with 55 patients with a left or right hemisphere middle cerebral artery stroke in which patients were assigned to a music group, a language group or a control group. Every day for 2 months, the music and the language groups listened to self-selected music or to audio-books, whereas the control group did not receive any listening material. Results showed that self-selected music listening during the early post-stroke stage could enhance verbal memory and focused attention even more than listening to audio-books. Verbal memory was evaluated thanks to a story recall subtest and a list-learning test whereas focused attention was assessed with mental subtractions and the Stroop test. No significant effect was found on the other cognitive domains. Furthermore, listening to music was indicated to prevent negative moods. In this study, 16 USN patients were included. However, their results were not analyzed separately from the other patients. In addition, the influence of music on spatial attention, and, especially on USN, was not precisely studied in this paper. How could music have a greater influence on cognition than stories? It is important to underline that some musics with lyrics were used in this study. Therefore, this material can provide more activation than the stories because the stories activate essentially the left cerebral hemisphere more dedicated to language whereas the combination of music and language can activate both cerebral hemispheres (Callan et al., 2006). Consequently, this can have a greater impact on cognition.

Additionally, in this study, music was shown to prevent depression. This suggests that music is closely linked to mood and emotions.

MUSIC EFFECT, AN EFFECT OF POSITIVE EMOTIONS INDUCED?

The question can arise regarding how music could enhance spatial attention in USN. Could the effects of music be specific to space or be linked to more general non-spatial effects, such as those induced by emotions?

The "Mozart effect" was the first piece of evidence in favor of interactions between music and space (Rauscher et al., 1993). This effect suggests that listening to Mozart's music can enhance visuo-spatial abilities. In their experiment, the authors found that a visuo-spatial quotient, calculated with series of tests assessing intellectual quotient, was higher for college students, just after listening to a Mozart's sonate. Rauscher et al. (1995) assumed that the same cerebral areas were activated by listening to a Mozart's sonate and when performing visuo-spatial tasks. However, this effect was not replicated by some studies (Steele et al., 1999). Moreover, Mehr et al. (2013) showed no cognitive effects of music instruction in children, suggesting that the "Mozart effect" is a temporary effect. Thus, a number of authors underlined that the "Mozart effect" could essentially be caused by a mood factor provoked by high tempo music (Thompson et al., 2001; Husain et al., 2002). This interpretation involving the influence of positive emotions induced by music has also been evoked by Soto et al. (2009).

Listening to pleasant music can influence emotional states and activate the substrates of emotional states in the orbito-frontal cortex (Dellacherie et al., 2009). According to Salimpoor et al. (2011), pleasant music produces a dopamine release in the mesocortico limbic reward system, precisely in the ventral and dorsal striatum. This increase of dopamine is at the origin of the activation of the orbito-frontal cortex. It also directly has an impact on other brain regions, such as ventral medial prefrontal cortex or hippocampus. Therefore, this dopamine release can enhance global cognitive functioning in patients with cognitive deficits (Nagaraja and Jayashree, 2001) as well as alertness, speed of information and memory in healthy individuals (Schuck et al., 2002).

Soto et al. (2009) hypothesized that these activated emotional regions could modulate the activity of intact attentional brain areas located in the intra-parietal cortex and, thus, increase visual awareness and, by the way, spatial attention.

Are these effects specific to music? Thompson et al. (2001) proposed the "arousal and mood hypothesis" that explains how cognition can be influenced by music. This hypothesis states that any enjoyable stimuli can induce positive affect and increase arousal and, consequently, improve cognitive performances. According to this hypothesis, this positive effect is not specific to music, it is essentially provoked by the emotional aspect of music. This interpretation was used by Sarkamo et al. (2008). Nevertheless, in neglect patients, another hypothesis could be assumed to explain why music could be useful to rehabilitate spatial attention. This hypothesis is based on more specific links between music and space and will be discussed in the next section.

LINKS BETWEEN SPACE AND MUSIC

Music could have a specific effect on the spatial attention of USN patients, not only induced by positive emotions. Space and music have close links. Some studies, carried out with healthy participants, showed that musicians had, on average, better visuo-spatial abilities than non-musicians. These studies have emphasized the idea of direct interactions between music and space. For example, the study by Brochard et al. (2004) indicated better performances in a perceptual and a mental visual imagery task in musicians than in non-musicians. Musicians were faster to associate a visual stimulus with a specific motor response. The authors assumed that daily practice of a musical instrument could have greatly improved basic perceptuomotor abilities, by reorganizing some cerebral networks.

The same cerebral areas, located in the parietal cortex are activated by tasks comparing different musical pitches and visuo-spatial tasks. Foster and Zatorre (2010) demonstrated the implication of the intra-parietal sulcus (IPS) in transforming musical pitch information. It should be noted that the IPS has a major role in space perception and in USN syndrome (Corbetta et al., 2005). Corbetta et al. (2000) also showed a hypo-activation of the IPS during spatial attention orienting task in all of their USN patients, indicating that there are close links between cerebral networks implicated in space attention and in music. Could music be a way to re-activate, or even reorganize, this specific network in the brain?

We can question why listening to musical tones can activate cerebral areas also related to space. The main considered hypothesis is that pitches can be conceptualized as positions on a mental line oriented either vertically or horizontally (Rusconi et al., 2006; Lidji et al., 2007). This hypothesis is inspired by the theory of the mental number line in which numbers are represented in a continuous analogical format (Dehaene, 2001). This means that numbers are ranked according to their magnitude on a mental line from left to right.

Zorzi et al. (2002) pointed out the fact that this mental number line could be disrupted in neglect patients. In this study, when USN patients had to estimate the midpoint of a numerical interval, a right-deviation in their answers was observed. These errors did not look like an acalculia but referred to their errors in bisecting lines. In addition, patients with right brain damage but without USN did not show this error pattern in this task. These findings suggested that numbers could have spatial representations.

According to some authors, numbers are not the only items linked to space. Walsh (2003) proposed A Theory Of Magnitude (ATOM) which suggests that space, numbers and other magnitudes are located in the inferior parietal cortex and share common representations. Buetti and Walsh (2009) have proposed that areas implicated by different sources of magnitude information are in the same cerebral region in order to improve the sensory-motor performance.

Lidji et al. (2007) carried out a series of experiments to prove the hypothesis of a mental pitch line. They showed using a Spatial Pitch Association Response Codes (SPARC) effect that a vertical line was activated automatically either in a pitch association task or in an instrumental timbre judgment task. This pattern of

results was observed in both musician and non-musician participants. In fact, if two buttons were vertically displayed, participants were faster to associate low-pitched tones with the bottom button and high-pitched tones with the top one. This vertical mapping of pitch is congruous with adjectives “low” and “high” used to describe the pitch of auditory stimuli. People associate pitch height with a spatial height.

The same authors also demonstrated the existence of a horizontal mental pitch line. The results were influenced by musical expertise of participants. Musicians seemed to associate automatically low-pitched tones with the left part of a horizontal line and high-pitched tones with the right part. For non-musicians, this association was found only with an explicit pitch comparison task and not with an instrumental timbre judgment task. These findings are congruent with those found by Rusconi et al. (2006) who carried out similar experiments on the SPARC effect.

Finally, Lidji et al. (2007) tested if an ascending or a descending melodic interval could activate either a left-to-right or a bottom-top association between spatial position and the orienting of the melodic interval change. No significant Spatial Melody Association Response Codes (SMARC) effect was found in this experiment in non-musicians. Participants were not faster to associate a descending melody with a left or bottom button and an ascending melody with a right or top button. Thus, melody was found to be too complex to be treated as spatial material. Overall, these findings indicate clearly that spatial areas are activated by a task using a pitch judgment and therefore could be activated by listening to music.

As USN patients could show impairment in the estimation of a number interval midpoint, we can ask ourselves if estimation of all magnitudes, and especially of pitches, could be affected by USN. Cusack et al. (2000) showed impairments in a pitch discrimination task in USN. Their patients had difficulties in localizing a sound relative to another in terms of frequency whereas no difficulty was found in one-interval task. These patients were able to say if a single sound was modulated in frequency. The authors concluded that their USN patients had a specific auditory deficit between-object comparisons and that within-object comparisons were entirely un-impaired. This study could be linked to those conducted by Lidji et al. (2007). USN patients have impairment in spatial cerebral areas, these impairments could affect the mental pitch line. The fact that one-interval tasks were well-performed could be compared with the fact that ascending or descending melodies do not imply a mental line. These tasks may involve other cerebral areas not impaired in USN.

Pavani et al. (2002) did not show difficulty in a pitch discrimination task. In this study, USN patients succeeded in discriminating a lower tone from a higher one similarly as a control group of patients with a right hemispheric lesion without USN. However, in this study, just two pitches were used, making the task too easy and not sensitive enough to detect a difficulty. Furthermore, USN patients had to categorize the sound they heard as “high” or “low” rather than localizing a pitch tone relative to another. This task could not imply spatial pitch representation. Therefore, pitch discrimination ability needs to be explored in USN with complementary studies.

How could these findings help to rehabilitate USN patients? Recently, Ishihara et al. (2013) have shown that listening to higher or lower pitches can modulate a line bisection task, either for healthy individuals or for right brain damaged patients with or without USN. A lower pitch induces a leftward or downward bias whereas a higher pitch involves a rightward or upward deviation. These effects were identified as greater only for the unique USN patient included in their study. In this patient, the bisection performance gradually increased during the experiment. Although only twenty lines were used in the experimental situation, this patient still showed an improvement on this task 1 week later. This study suggests that non-lateralized auditory cues, especially pitches, could influence the direction of the attentional bias in USN. The authors have underlined the fact that these auditory cues could be used as a long-lasting rehabilitative treatment. Thus, one more time, more researches are needed to assess the efficiency of using pitches as spatial cues in a USN rehabilitation.

Although pitch discrimination has not been studied enough in USN, evidence seems to show that music could activate spatial representations in the brain and, therefore, could be a successful tool to use in the rehabilitation of USN.

MUSICAL PRACTICE: A PROMISING TOOL TO REHABILITATE USN?

Musical practice appears to be a promising tool to rehabilitate USN for several reasons. First, playing music involves several sensory modalities: hearing, vision, touch and motor skills. Musical practice also implies higher-order cognitive processes. Musicians learn associations between motor actions, specific sound and visual stimuli and receive, in return, multisensory feedback. According to Wan and Schlaug (2010), these associations can strengthen connections between auditory and motor regions and activate multimodal integrations regions around the IPS, which is hypo-activated in USN (Corbetta et al., 2000). In addition, as mentioned earlier, the fact that music could activate spatial representations in brain has been stressed either for musicians or for non-musicians (Lidji et al., 2007). Therefore, playing music could be an interesting way to re-activate brain networks impacted by USN.

Finally, playing music could have a greater impact than other methods. USN patients can have difficulties in being implicated in their own rehabilitation, in particular because of anosognosia (i.e., unawareness of the neurological deficit). Appelros et al. (2007) estimated that anosognosia touched 43% of USN patients and partly explained why rehabilitation fails for some patients. Musical practice may be more pleasant than commonly used rehabilitation methods and, therefore, could be less challenging for patients who are not aware of their difficulties.

The real efficiency of musical practice has not been sufficiently examined in USN. In the literature, just one study, in which only two USN patients were included, implied musical practice (Bodak et al., 2014). The authors showed that an active period of music-making with a horizontally aligned instrument (chime bars) could reduce attentional bias. Unfortunately, only conventional tasks were used to evaluate USN in this study (cancellation tasks, line bisection, ...) and no ecological test was included and, therefore, we cannot conclude on potential daily life generalization.

Furthermore, this study did not include another rehabilitation method to compare the efficiency of musical practice and so the observed improvement could only be an effect due to the introduction of a retraining and not a specific effect as it was expected to prove. Clearly, more research is necessary in order to fill the theoretical and applicative gaps found in the literature concerning music and, more particularly, musical practice.

CONCLUSION

Several sensory rehabilitation techniques have been proposed for USN patients. As we saw in this review, a number of them focus only on the visual modality whereas others use motor skills, somato-sensory or vestibular stimulations to improve visual spatial attention. These rehabilitation methods are mainly based on two theoretical approaches: either recalibrating cognitive maps or retraining the orientation of spatial attention. The literature indicates that poly-sensory rehabilitation appears to be a better way to rehabilitate USN. However, all these methods present major issues concerning the generalization to daily life and the long-term efficiency.

In the light of this review, both music listening (perception) and music playing (production) have been indicated as promising methods to rehabilitate spatial attention in USN patients. Music involves general effects on cognition linked to motivation and the emotions that it induces (the heightened arousal produced by music has an impact on cognition). We have seen that music could contribute more specifically to the orientation of spatial attention as music and space share closed links. Notably, musical practice could be an interesting rehabilitation tool as it also involves several sensory modalities (hearing, vision, arm movements, etc). It is essential that more research is conducted on music and musical practice in order to determine their potential effects on spatial attention in USN patients.

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New framework for rehabilitation – fusion of cognitive and physical rehabilitation: the hope for dancing

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Neurorehabilitation programs are commonly employed with the goal to help restore functionality in patients. However, many of these therapies report only having a small impact. In response to the need for more effective and innovative approaches, rehabilitative methods that take advantage of the neuroplastic properties of the brain have been used to aid with both physical and cognitive impairments. In line with this, there has been a particular interest in the use of physical exercise as well as musical related activities. Although such therapies demonstrate potential, they also have limitations that may affect their use, calling for further exploration. Here, we propose dance as a potential parallel to physical and music therapies. Dance may be able to aid with both physical and cognitive impairments, particularly due to its combined nature of including both physical and cognitive stimulation. Not only does it incorporate physical and motor skill related activities, but it can also engage various cognitive functions such as perception, emotion, and memory, all while being done in an enriched environment. Other more practical benefits, such as promoting adherence due to being enjoyable, are also discussed, along with the current literature on the application of dance as an intervention tool, as well as future directions required to evaluate the potential of dance as an alternative therapy in neurorehabilitation.

Keywords: neurorehabilitation, dance, combined therapy, plasticity, music therapy

INTRODUCTION

Neurological disorders have been estimated to affect as many as a billion people worldwide, with this number expected to increase in the upcoming years (World Health Organization, 2006). Such disorders can be heterogeneous in regards to their symptoms and can include any combination of impairments related to physical and cognitive functioning, or issues with behavior, all of which can impact the basic daily living capability of individuals. Unfortunately, there are no current treatments that can address all symptoms in a meaningful manner. Surgical and pharmacological therapies for prevalent neurological disorders, such as Alzheimer's and Parkinson's, have been developed, but may only address a subset of symptoms, and even then, do so with limited efficacy (Ahlskog, 2011; Ahlskog et al., 2011; Intlekofer and Cotman, 2013). Adjunct conventional rehabilitative programs have been used as part of treatment regimes, although the effectiveness of such conventional therapies may also be limited (Lincoln et al., 1999; Langhammer and Stanghelle, 2000; Woldag and Hummelsheim, 2002; Bassett, 2003).

Recently, scientists have highlighted that exercise and music related activities can induce neuroplasticity, and are capable of aiding with physical and cognitive impairments across various neurological patient groups, including in those suffering from dementia and Alzheimer's (Heyn et al., 2004; Thompson et al., 2005; Irish et al., 2006; Bruer et al., 2007; Lautenschlager et al., 2008), stroke (Duncan et al., 1998, 2003; Gordon et al., 2004; Schneider et al., 2007; Särkämö et al., 2008; Quaney et al., 2009), and Parkinson's

disease (Thaut et al., 1996; Crizzle and Newhouse, 2006; Goodwin et al., 2008; Tanaka et al., 2009; Cruise et al., 2011). However, the uses of such therapies are also faced with inherent limitations, creating the need to explore for further options.

Here, we propose dance as an intriguing alternative to physical and musical therapies as used in neurorehabilitation. As a physical activity, dance may be able to aid with physical functioning. However, other elements found in dance may also contribute to it being a cognitively stimulating activity. This may allow dance to have a positive impact on not only physical functioning, but cognitive as well, in part due to fitting the framework of what are known as combined, or multimodal, therapies, which incorporate simultaneous physical and cognitive activity in a stimulating environment (Lustig et al., 2009; Kraft, 2012). Dance may also be able to overcome some of the more practical limitations associated with other alternative therapies. In the following sections, the current literature on the use of physical and musical activities in neurorehabilitation is discussed, along with the limitations associated with such therapies, and how dance can be a potential alternative.

PHYSICAL EXERCISE AND NEUROREHABILITATION

Physical activity, particularly aerobic exercise, has recently drawn interest for its potential use in neurorehabilitation. Engagement in physical exercise has been commonly reported as being associated with a reduction in risk for various neurological disorders, notably for cognitive decline, dementia and Alzheimer's (Larson et al., 2006; Hamer and Chida, 2009; Sofi et al., 2011; Buchman et al.,

2012). There is also support linking engagement in physical exercise to a reduced risk for onset of Parkinson's (Xu et al., 2010) as well as stroke incidence (Do Lee et al., 2003), although these findings are not as robust as those for dementia and cognitive decline. Such epidemiological related studies suggest exercise having a neuroprotective effect in relation to the onset of various neurological disorders (Hillman et al., 2008). However, these findings do not offer support for how physical exercise can be used as an intervention for those who already suffer from such disorders. Rather, the interest in using exercise for neurorehabilitation stems from training based animal and human studies, which have helped demonstrate the widespread impact exercise can have on cognition, plasticity and overall brain health, particularly on the aging brain.

Although not directly translatable to humans, studies in animals have helped elucidate the fine neural changes that occur in the brain as a result of exercise. The impact of exercise on neurogenesis has been one of the focal points of such studies. Although a decline in neurogenesis is associated with aging, one of the most consistent findings in animal models is an increase in neurogenesis in the dentate gyrus of the hippocampus as a result of exercise (Hillman et al., 2008; van Praag, 2008), which is associated with improvements in cognition, particularly in learning and memory (van Praag et al., 1999, 2005; Vaynman et al., 2004; van Praag, 2009). As for the clinical significance, it has been hypothesized that a reduction in neurogenesis, as found in Alzheimer's (Hillman et al., 2008), may exacerbate memory impairments, possibly through interfering with hippocampal neural circuits (Lazarov et al., 2010); exercise may represent an endogenous approach for the restoration of cells in the hippocampal dentate gyrus. However, the impact of exercise on neurogenesis and how this relates to improving cognitive impairments in a clinical context remains unclear (van Praag, 2008, 2009; Lazarov et al., 2010; Mu and Gage, 2011).

Exercise can also affect brain vasculature, particularly by increasing angiogenesis throughout the brain (Cotman et al., 2007; Voss et al., 2013), including in regions such as the hippocampus (Kramer and Erickson, 2007), motor cortex (Kleim et al., 2002; Swain et al., 2003), and cerebellum (Black et al., 1990). Brain health can be impacted via new blood vessels being used to deliver necessary nutrients to both new and old neurons in the brain (Kramer and Erickson, 2007). Neurotrophic factors, which promote plasticity, such as insulin-like growth factor, fibroblast growth factor 2, and brain derived neurotrophic factors (BDNFs), have also been shown to be upregulated following exercise treatments (Cotman and Berchtold, 2002; Vaynman and Gomez-Pinilla, 2005). BDNF has been of particular interest due to its potential role in promoting neural reorganization and regeneration in an impaired central nervous system (Vaynman and Gomez-Pinilla, 2005). Its impact on neurorehabilitation may stem from its crucial role in hippocampal learning and memory formation through long term potentiation (Egan et al., 2003; Hillman et al., 2008), its involvement in neuroprotection and promotion of cell survival (Kramer and Erickson, 2007), as well as recovery of motor functions (Griesbach et al., 2004; Ploughman et al., 2009).

Intervention based human studies, with a focus on healthy older adults, have also been able to show that exercise training

can enhance brain plasticity and cognition. In a meta-analysis, Colcombe and Kramer (2003) surveyed 18 studies that included randomized aerobic fitness training in older adults, and reported a moderate effect size for the impact of fitness training on cognition. Specifically, greater executive, controlled, spatial and speed processes were linked to fitness training, with executive control showing the largest effect size. Intervention based neuroimaging studies in healthy adults have also shown that physical exercise can lead to various changes in the brain, including increased functional activity in frontal and parietal regions and decreased functional activity in anterior cingulate cortex regions (Colcombe et al., 2004), increased gray matter volume in regions of the frontal and superior temporal lobe (Colcombe et al., 2006) as well as in the hippocampus (Erickson et al., 2011), and significant increases in the blood volume of the hippocampal dentate gyrus (Pereira et al., 2007). Such changes, particularly those in the hippocampus, have also been found to be correlated to improvements in cognition (Pereira et al., 2007; Erickson et al., 2011). Thus, both animal and human studies have helped elucidate the various ways in which exercise can have an affect on the brain.

In regards to application to neurological groups, exercise has been found to aid with physical and functional impairments, including in stroke (Duncan et al., 1998, 2003; Gordon et al., 2004), Parkinson's (Crizzle and Newhouse, 2006; Goodwin et al., 2008), and dementia patients (Heyn et al., 2004). Some studies suggest that exercise can also improve cognitive functioning in neurological groups (Heyn et al., 2004; Quaney et al., 2009; Tanaka et al., 2009; Baker et al., 2010; Cruise et al., 2011; Nagamatsu et al., 2013), although the current evidence has also been viewed as being mixed (Angevaren et al., 2008; Forbes et al., 2008; Busse et al., 2009; McDonnell et al., 2011; Snowden et al., 2011). Reports of small effect sizes (Lautenschlager et al., 2008), gender effects (Baker et al., 2010), variance in what cognitive domains are reported to be improved (Angevaren et al., 2008), as well as methodological differences (McDonnell et al., 2011; Snowden et al., 2011), are current limitations as reported in the literature, making it difficult to interpret the efficacy of exercise in neurorehabilitation.

There may also be limitations associated with transferring the use of exercise to real world clinical applications. For example, some of the studies discussed used moderate to high intensity exercises (Lautenschlager et al., 2008; Baker et al., 2010; Cruise et al., 2011), although those with more severe symptoms may not be able to be active at such levels (Snowden et al., 2011). Also, adherence issues may impact the use of conventional exercise as a therapy. It has been estimated that over 50% of participants who begin an exercise program will drop out within the first 6 months (Dishman, 1988). This is particularly prominent in older adults, who may initially be willing to participate in an exercise program, but only do so for the short term, eventually stopping (Van Der Bij et al., 2002). Unfortunately, the benefits of exercise require continued participation, making the issue of adherence that much more important. Given that neurological disorders affect patients for the long term, it is important that any therapy used is capable of promoting continuous commitment that ends up becoming part of the person's weekly routine. Various factors that may contribute to whether someone will continuously

participate in an exercise program include social support, health, personal beliefs, as well as motivation and enjoyment (Rhodes et al., 1999). However, traditional exercise programs may not fulfill many of these needs; in particular, conventional methods of exercise may not be seen as enjoyable by participants, which can have a significant impact on adherence (Rhodes et al., 1999; Belardinelli et al., 2008; Findorff et al., 2009). Such issues are important to keep in mind when promoting the use of physical exercise for neurorehabilitation.

In summary, there is strong support from both animal and human studies that physical exercise can be beneficial for the brain. It is important to note that the vast majority of these studies are based on the use of exercise in healthy populations, where a major focus has been on its potential role in healthy aging. Unfortunately, the efficacy of exercise for aiding with cognition in neurological disorders remains difficult to interpret. Both animal and human clinical studies have yet to produce the robust findings of the impact of exercise as found in their healthy counterparts. Methodological differences found between studies also contribute to the effects of exercise being unclear. Other more practical factors, such as the intensity of exercise needed to produce meaningful results, as well as enjoyment and adherence issues, also pose potential limitations for the application of exercise in neurorehabilitation.

MUSICAL ACTIVITIES AND NEUROREHABILITATION

Music has been described as a powerful multimodal stimulus in humans (Sacks, 2006), invoking the widespread activity of various brain regions related to sensorimotor, higher order cognitive and emotional processes (Menon and Levitin, 2005; Koelsch, 2009; Herholz and Zatorre, 2012). Such processes can include auditory processing, attention, memory and sensory-motor integration, leading to the involvement of networks that consist of frontal, temporal, parietal and subcortical regions (Zatorre, 2005; Schlaug, 2009). Music processing can also be quite a complex task, recruiting various brain regions that are associated with the different components found in music, including pitch, timbre, rhythm, melody, recognition, and emotion (Lin et al., 2011). Musical experiences have also been shown to extensively enhance brain plasticity across the lifespan, leading to alterations of structural and functional properties of the human brain (Gaser and Schlaug, 2003; Schlaug et al., 2005; Hyde et al., 2009; Herholz and Zatorre, 2012). The ability of music to stimulate the widespread activity of brain regions through the engagement of various processes, as well as being able to enhance neuroplasticity, has led to interest in how the effects of musical activities can be generalized beyond the musical domain, and be used to improve various neurological-related impairments (Schlaug, 2009; Thaut et al., 2009; Altenmüller and Schlaug, 2013; Hegde, 2014).

Engaging in musical activities has been found to have a powerful effect on cognitive functions across the lifespan. Although aging is associated with a decline in various cognitive and perceptual domains (Rowe and Kahn, 1997), engagement in music related activities at a younger age may be able to help mitigate some of these effects later on in life. For example, in older adults, prior musical experience has been associated with enhanced perception of speech in a noisy environment as well as auditory working memory capacity (Parbery-Clark et al., 2011), a delay in the age

associated decline of neural encoding of speech perception in the brainstem (Parbery-Clark et al., 2012), as well as a greater preservation of cognitive functioning in domains such as non-verbal memory and executive processes (Hanna-Pladdy and MacKay, 2011). Also, although normal aging is associated with a decrease in gray matter volume (Good et al., 2001), musical training throughout life may be related to the prevention of such decline (Sluming et al., 2002). However, the benefits of music do not strictly apply to only those who have engaged in musical activities throughout their life. Instead, musical training has also been found to be a powerful intervention tool for improving cognition in healthy older adults (Bugos et al., 2007). Simply listening to music has also been shown to enhance cognition in both young (Thompson et al., 2001; Schellenberg et al., 2007) and older adults (Thompson et al., 2005), although such findings are attributable to the enhancement of arousal and mood, rather than music itself. With music being a powerful stimulus, music based activities have been viewed as holding potential for being an engaging and effective method of neurorehabilitation (Schlaug, 2009).

Musical therapies, in both active and passive forms, have been used in neurorehabilitation. For example, training in the playing of instruments has been used to improve motor movements, including range, speed and quality of movements, in stroke patients (Schneider et al., 2007). Such improvements were also found to be associated with increased activity of motor regions, as well as neural reorganization of the motor network (Altenmüller et al., 2009). The coupling of auditory-motor stimulation found in playing instruments may have a role in recovery of motor skills, with improvements possibly being associated with an increase in functional auditory-motor connectivity (Rodríguez-Fornells et al., 2012). The relationship between motor movement and auditory stimuli has also been the basis of rhythmic auditory stimulation, which consists of movements being done to rhythmic auditory cues, and has been found to lead to functional improvements in patients with brain injuries (Hurt et al., 1998) and Parkinson's (Thaut et al., 1996), with it being particularly effective in aiding with gait and upper extremity functioning (Thaut and Abiru, 2010). Music therapies have also been developed for speech and language impairments, such as Melodic Intonation Therapy, which has been used to improve speech in non-fluent aphasics by having patients sing phrases in a manner that exaggerate the melodic content of normal speech, all while tapping in synchrony to the syllables with their left hand (Schlaug et al., 2010). Preliminary findings from general music therapies, which consist of participating in various musical related activities, have also shown to improve executive functioning in those with brain injuries (Thaut et al., 2009). Thus, various types of musical therapies have been shown to be of use in different clinical groups.

Listening to music can lead to the engagement of various sensorimotor, cognitive and emotional processes in the brain (Zatorre, 2005; Koelsch, 2009), invoking the widespread activity of temporal, frontal, parietal, subcortical, and cerebellar regions (Särkämö and Soto, 2012). With music listening being an engaging and cognitively stimulating activity, it has also been explored as a potential tool for neurorehabilitation. Särkämö et al. (2008) investigated the effects of passive music exposure on cognition and

mood in stroke patients. Patients were placed into a music listening group, a language group, or a control group, which consisted of listening to self-selected music, audio books or nothing, respectively. Music listening was found to enhance cognitive recovery, providing greater improvements in focused attention and verbal memory, in comparison to the language and control group. Improvements were also noted for depression and mood as a result of music listening. However, whether such effects were specific to music, were primarily influenced by arousal, or can have a long lasting impact remains unclear (Särkämö et al., 2008). A more recent study by Särkämö et al. (2014) showed that listening to music can lead to structural changes in the brain of stroke patients, particularly leading to an increase of gray matter in frontolimbic brain areas; these structural alterations were also found to be correlated with improvements in cognition. Music listening has also been associated with improvements in visual awareness and attention in patients with neglect (Soto et al., 2009; Tsai et al., 2013), aiding with memory in those with Alzheimer's (Simmons-Stern et al., 2010), as well as improving cognitive performance in those with dementia related impairments (Thompson et al., 2005; Irish et al., 2006; Bruer et al., 2007).

Although there is no clear answer as to how music listening can provide therapeutic value, certain mechanisms have been proposed to be of potential importance. Music has been shown to be a powerful emotional stimulus, capable of modulating emotional and reward related systems (Blood and Zatorre, 2001; Brown et al., 2004; Menon and Levitin, 2005). This effect may be able to improve mood and arousal in patients, which may in turn positively impact how they react to therapy and rehabilitation (Van de Winckel et al., 2004). Listening to pleasurable music may also be able to enhance cognitive functioning through modulating the release of dopamine (Särkämö and Soto, 2012), which has been inferred based on dopamine release being associated with emotional arousal during musical listening (Salimpoor et al., 2011), and the increase of dopamine being associated with various improvements in cognition (Husain and Mehta, 2011). The potential long term effects of music listening may be related to how it may enhance neuroplasticity, which may include promoting neurogenesis in the hippocampus (Kim et al., 2006), increasing BDNF levels in the hypothalamus (Angelucci et al., 2007a) and hippocampus (Angelucci et al., 2007b), as well as providing an auditory enriched environment which can positively impact auditory cortical functioning (Engineer et al., 2004). It has also been speculated that listening to music can enhance the regeneration and repair of neurons through influencing the secretion of specific steroid hormones (Fukui and Toyoshima, 2008). Thus, although arguably the simplest form of a musical experience, just the act of listening to music may have a powerful influence.

Other properties of music which may also further enhance its therapeutic value include its potential influence on neurochemical changes across various domains, including stress and arousal (Chanda and Levitin, 2013), or how musical therapies can be seen as enjoyable activities, in part due to the presence of music, which can improve mood and motivation, and thus the efficacy of interventions (Schneider et al., 2007; Herholz and Zatorre, 2012). However, there are limitations for the application

of music related activities in neurorehabilitation. Passive music therapies when applied to groups with cognitive or behavioral issues may offer little support to improve general physical functioning, which although may not be the primary symptoms, can still be affected. Active musical therapies may aid with physical symptoms, but they are often reported as being used to assist with very specific motor impairments, tailored to the needs of the patient, and lacking any impact on broader general functioning. The efficacy of musical activities, particularly that of passive listening, in aiding with cognitive impairments also remains unclear, with methodological issues in current studies cited as making it difficult for any clear-cut conclusions to be drawn (Vink et al., 2003). There also may be more practical issues, such as hearing impairments amongst the elderly (Gordon-Salant, 2005), which may impact therapies that require music listening, or the issue of using instruments, and expecting patients to learn to play in active musical training therapies. There are also questions about the long-term impact of music listening on cognition, and the specific cognitive domains music listening may be able to aid with (Särkämö et al., 2008). That is not say that listening to music cannot improve cognition, but rather there is a gap between the understanding of music's potential role in improving cognitive impairments, and the existence of meaningful results in the widespread application of music related activities to various neurological populations (Hegde, 2014). Thus, although musical therapies have provided promising results, their use as a neurorehabilitative tool for aiding with various neurological-related symptoms, particularly that of cognitive impairments, requires further investigation.

DANCE AND NEUROREHABILITATION

WHAT IS DANCE

Dance may be defined as the act of one or more bodies moving in a rhythmic manner cued by music. However, even in its simplest form, dance requires a complex and simultaneous engagement of various physical and cognitive faculties. The specific elements of dancing can vary greatly, but common features include learning new sequences of movements and rehearsing them, music accompaniment, and typically being held in a group, fostering social interaction. Comparisons between expert dancers and controls have also helped demonstrate the various training effects associated with dancing, including training effects related to not only physical fitness, such as posture control (Simmons, 2005; Rein et al., 2011) and balance (Crotts et al., 1996; Gerbino et al., 2007; Bruyneel et al., 2010), but cognition as well, including in specific memory domains and tasks (Starkes et al., 1987; Smyth and Pendleton, 1994; Hübner et al., 2011). Expert dancers have also been found to have structural differences in sensorimotor networks (Hänggi et al., 2010) and the hippocampus (Hübner et al., 2011), as well as functional neural differences related to their training, such as when visualizing movement to familiar versus unfamiliar music (Olshansky et al., 2014), supporting the notion that training in dance can induce specific neuroplastic changes. This ability for dance to be both a physical and cognitive engaging activity may hold value for its potential as an alternative therapy for various clinical groups. The use of dance for aiding with physical functioning will be discussed, but here we focus on a lesser

explored topic, that of which how dance may also be able to aid with cognitive functioning.

INTEREST IN DANCE FOR NEUROREHABILITATION

Combined training

Physical and cognitive training on their own have been shown to be useful to some extent for improving cognition, but there may be added benefits to combining the two into a single activity. It has been proposed that exercise may need to be done in a cognitively stimulating context in order to maximize its impact on neuroplasticity and cognition (Fabel and Kempermann, 2008). In regards to how such combined interventions can lead to greater functional benefits than physical or cognitive activity alone, animal studies suggest a synergistic effect, possibly due to differences in how physical and cognitive activity induce neuroplasticity. For example, both physical and cognitive activities have been shown to increase neurogenesis in the hippocampus (Kempermann et al., 1997; van Praag et al., 1999), as well as improve learning and memory for hippocampal related tasks (Olson et al., 2006). However, the way in which they increase neurogenesis may be quite distinct, with exercise leading to an increase in neurogenesis by increasing precursor cell proliferation, whereas an enriched environment promotes the survival of new cells (Kempermann et al., 2010). Fabel et al. (2009) found that in mice, physical exercise followed by cognitive stimulation through an enriched environment resulted in an additive effect on neurogenesis. It is thought that although physical exercise can increase the precursor cell pool, cognitive stimulation may increase the recruitment of cells to be integrated into functional networks (Fabel and Kempermann, 2008). Animal studies also show support for combined physical and cognitive activities being able to significantly enhance learning and working memory abilities compared to either activity done alone, independent of exercise intensity or duration (Langdon and Corbett, 2012).

Although the literature for the explicit use of combined training in humans is limited, results from these studies are nonetheless promising. Combined training programs have been reported as providing significantly greater improvements in older adults across various cognitive functions compared to isolated physical and cognitive training programs (Fabre et al., 2002; Oswald et al., 2006). A more recent study by Anderson-Hanley et al. (2012) aimed at comparing the effects of physical exercise in a standard environment versus that in a cognitively stimulating environment. The study consisted of participants being placed into either a standard group, which consisted of stationary cycling in a traditional fashion, or in an experimental group, which consisted of stationary cycling with a virtual reality display. Although participants did not engage in any explicit cognitive tasks, the use of virtual reality in the context of exercise may be seen as cognitively stimulating since it would putatively recruit additional neural networks. Executive functioning was found to be significantly better in the virtual reality group compared to the standard group. Those in the virtual reality group were also found to have a significantly greater increase in BDNF levels. However, exercise related effort and fitness between the two groups were similar, suggesting that not physical exercise alone, but rather the combined physical and cognitive activity, led to the greater improvements in cognition

found in the experimental group. Combined therapies have also been found to be effective in improving cognition in neurological groups (Suzuki et al., 2012; Coelho et al., 2013), although it is important to note that such studies only compared the combined group to a control group, making it impossible to compare the effects of the combined therapy to physical or cognitive only activities. Thus, although further investigation is required, preliminary findings from both human and animal studies suggest that there may indeed be additional benefits to combined therapies.

Dance as a combined intervention

Dancing may be considered an ideal example of what multimodal training should consist of, due to combining physical and cognitive activity together (Fissler et al., 2012; Kraft, 2012; Olsson, 2012) in an enriched environment (Kattenstroth et al., 2010). Aside from the physical activity that dancing requires, it also involves various cognitive functions such as perception, emotion, executive functioning, memory and motor skills (Foster, 2013). Indeed, neuroimaging studies have shown widespread activity in the brain during rehearsal of dance movements (Brown et al., 2006). The engagement of such a wide variety of cognitive faculties may be attributed to dance being an experience that provides multisensory stimulation in an engaging environment, due in part to incorporating components such as physical activity, music listening and social interaction (Johansson, 2012). How dance engages various cognitive faculties may be best answered by examining the effects of dance training on experts.

As previously described, training in dance requires substantial physical and cognitive engagement. Dancing consists of being aware of one's appropriate sequence of movements over time, as well as how these movements should be conducted in relation to external cues (Sevdalis and Keller, 2011). This may be evident by training effects found in expert dancers in comparison to non-dancers for various cognitive tasks, including experts showing a greater recall for structured sequences (Starkes et al., 1987), longer memory spans for ballet and nonsense movements (Smyth and Pendleton, 1994), and significantly greater performances on non-spatial memory tasks (Hüfner et al., 2011). Learning sequences of movements may also be enhanced in a dance setting through observation of dynamic kinematic information provided from other dancers during learning (Gray et al., 1991). Brain activity in response to action observation has also been implicated in dancing, with experts being shown to have an increase in activity in brain regions considered a part of the human mirror system when viewing well known movements in relation to their learning of dance (Calvo-Merino et al., 2005; Cross et al., 2006). Also, the neural substrate of learning to dance through effective observation may be similar to that of learning by physical practice (Cross et al., 2009), with speculation that the effective observational learning associated with dance may influence other cognitive skills as well (Kattenstroth et al., 2013). Other factors that may also enhance the ability of dance to be a cognitively stimulating activity may include its social nature. The social environment dancing provides may be a source of cognitive stimulation, where simple exercises may recruit a more widespread brain network if done with other people in comparison to alone (Saarela and Hari, 2008). However,

two other sources of substantial cognitive stimulation in dance include the musical element it incorporates, as well as the complex physical and motor skills it requires, both of which will be expanded upon.

Music in dance. Dance is undoubtedly a complex task, made of numerous components that come together in sequence. However, a particular feature of dance that may bolster its therapeutic value is its incorporation of music. As already discussed, music is a powerful source for auditory stimulation with an ability to engage numerous emotional and cognitive faculties (Menon and Levitin, 2005; Peretz and Zatorre, 2005), and it may be speculated that the simple exposure to music may play a big role in dance being a cognitively stimulating activity. However, in combined interventions, cognitive and physical stimulation are seen as having an interaction effect, where the benefits gained from combining the two outweigh the benefits from either activity done alone. Thus, in the context of dance as a combined therapy, it is important to address whether music can enhance the benefits already found in physical activity.

Exercise conducted with music has been shown to lead to significant improvements in cognition in various clinical groups (Emery et al., 2003; Van de Winckel et al., 2004), but whether physical activity with music can lead to greater improvements than exercise alone is still not clear. However, a recent study by Satoh et al. (2014) seems to suggest that there may be indeed an additive effect on cognition when the two are combined. Satoh et al. (2014) placed older healthy adults into either a control group, an exercise group, or an exercise with musical accompaniment group. The physical routine between the two exercise groups was identical, with the only difference being the musical element found in the combined group. Cognitive improvements were greatest in the combined music and exercise group, with visuospatial functioning found to be significantly greater in the combined group than in the exercise alone group. It was suggested that the findings may be explained in the multimodal framework, where the addition of music to exercise could have led to simultaneous physical and cognitive stimulation, producing greater cognitive improvements than exercise alone. Such findings may give credence to music alone being able to provide enough cognitive stimulation where it is able to significantly enhance the effects of exercise. These findings may also be seen as providing additional support that dance can be a method of multimodal stimulation. Aside from passively listening to music, the act of timing and synchronizing movements to music may also be cognitively demanding (Bläsing et al., 2012). In comparison to the lack of music, dancing with music has been shown to activate unique regions including the anterior vermis of the cerebellum, putamen and the medial geniculate cortex, regions believed to be involved in synchronizing movement to musical rhythms during dance (Brown and Parsons, 2008). Dancing may also be viewed as a dual tasking activity, with the need for attention to be divided amongst various components, such as navigation and balance (Hackney et al., 2007); music may also play a factor in this. In contrast to listening to a piece of music, which can consist of a state of undivided attention, dance requires not only the listening and processing of music, but also moving accordingly to the music being listened to, which may present another form of

dual tasking. Indeed, musicians have been shown to outperform non-musicians on dual task performance, potentially in part due to the fine motor coordination musicians require while simultaneously processing various musical elements (Moradzadeh et al., 2014). Dance, although perhaps to a lesser extent, can also be seen as being an exercise in dual tasking for similar reasons, due to requiring the fine physical and motor coordination of movements being done to music. As previously described, the impact of physical activity on cognition may be greatest when done with complex cognitive stimulation (Fabel and Kempermann, 2008). Music may act as such a source of stimulation, with its ability to engage various cognitive faculties and induce widespread activity throughout the brain (Zatorre, 2005; Schlaug, 2009). Thus, when portraying dance as a combined therapy, its incorporation of music may be just one, albeit very powerful, source of cognitive stimulation.

Physical and motor skills in dance. In a multimodal framework, physical activity is used to place the brain in a plastic state, preparing the brain to respond to cognitive stimulation, and thus allowing for a greater impact of cognitive activities (Hotting and Röder, 2013). However, the physical activity itself may also be a source of cognitive stimulation. The question then becomes what physical activities should be used in combined therapies. Aerobic exercises, such as running, are a popular choice, but due to some of the more complex movements, as well as the learning that is required for such movements, dance may offer a unique method of physical exercise to induce neuroplasticity and enhance cognition.

More conventional aerobic exercises, such as walking, have been used with success in multimodal training (Fabre et al., 2002; Anderson-Hanley et al., 2012; Suzuki et al., 2012; Coelho et al., 2013). Changes to cardiorespiratory fitness, as a function of aerobic exercise intensity, are believed to contribute to aerobic exercise being able to provide improvements in cognition (Hayes et al., 2013). However, dance may not hold the same intensity as other aerobic exercises, particularly for older adults. For example, Kattenstroth et al. (2013) reported no improvement in cardiorespiratory performance in older healthy adults after attending weekly dance classes for 6 months. This was attributed to the low intensity nature of the dance class, which consisted of many disruptions during dancing to provide new instructions, thus preventing any continuous activity to take place for an extended period of time. Yet, even if low intensity dancing is not able to improve cardiorespiratory performance in older adults, it may still be able to positively affect cognition. It has been noted that results from the literature do not support the notion that exercise can lead to improvements in cognition only if cardiovascular fitness is significantly impacted (Etnier et al., 2006; Busse et al., 2009). Rather, physical activity, even at a low intensity where there is no significant change to cardiorespiratory fitness, may still be able to positively affect cognition and brain structure (Ruscheweyh et al., 2011; Hayes et al., 2013). Low intensity exercise has also been noted to still be able to enhance neuroplasticity, for example, increasing BDNF levels (Soya et al., 2007; Aguiar et al., 2011; Vaughan et al., 2014). The ability for low intensity activity to still have a positive impact on the brain may help explain why Kattenstroth et al. (2013) found no changes to cardiorespiratory fitness, but still reported significant

improvements in various cognitive and sensorimotor domains as a result of dancing. That is not to say that dancing is incapable of leading to significant changes in cardiorespiratory fitness, since it has been shown to do so (Hopkins et al., 1990). Rather, low intensity dancing, regardless of whether it provides significant changes in cardiorespiratory fitness, may still be able to positively influence brain plasticity and cognition in a similar manner to other aerobic exercises. Yet, there may be other unique properties of dancing not found in other conventional aerobic exercises that may lead it to having a greater affect on cognition.

Dance can be viewed as a multifaceted activity, including not only physical fitness, but also motor fitness, which accounts for components such as balance, flexibility, speed and coordination (Voelcker-Rehage et al., 2010). The extensive engagement in motor fitness may be evident by trained dancers having been shown to be significantly better than non-dancers across a variety of motor fitness domains, including posture control (Simmons, 2005; Rein et al., 2011) and balance (Crotts et al., 1996; Gerbino et al., 2007; Bruyneel et al., 2010). Expert dancers have also been shown to have a structurally altered sensorimotor network, including regions such as the premotor cortex, supplementary motor area and putamen (Hänggi et al., 2010). These alterations have been speculated to be related to dancing requiring intensive training in motor skills such as precise posture control, as well as coordination of the body for both gross and fine motor movements (Hänggi et al., 2010). Dancing has also been found to elicit significant improvements in various domains of motor fitness in older adults (Hopkins et al., 1990; Shigematsu et al., 2002; Verghese, 2006; Keogh et al., 2009; Kattenstroth et al., 2013). There is also evidence to suggest that both physical and motor fitness in older adults are positively associated with cognition, although the two may differ with respect to the specific cognitive domains that they are associated with (Voelcker-Rehage et al., 2010). Exercises that combine physical and motor fitness have also been found to lead to significant improvements across a variety of cognitive domains in older adults (Vaughan et al., 2014). Coordination related exercises have been shown to require greater attention and concentration during execution compared to simple physical tasks (Mochizuki and Kirino, 2008). These findings do not necessarily suggest that one of motor and physical fitness exercises is in a sense better than the other. Rather, each may provide its own unique benefits, leading to the speculation that exercises that combine both may have a greater impact on cognition than those that only involve either type of fitness. In this context, dance may be an ideal activity due to requiring participants to extensively engage in both physical and motor fitness, and thus may be more encompassing in regards to engaging different cognitive domains in comparison to other conventional exercises.

The difficulty of a physical activity and its requirement for novices to learn new physical skills may also be a factor in the potential neuroplastic changes induced by dance. Black et al. (1990) found that in rats, acrobatic training led to greater synaptogenesis in the cerebellar cortex than simple physical exercise training. This was attributed to the acrobatic training consisting of learning new and difficult motor tasks, whereas the physical exercise consisted of simple and repetitive activity of an already well known task (walking). Similar findings were reported by

Curlik et al. (2011), who investigated the effects of physical activity that require skill learning on the survival of cells in the adult hippocampus. Rats were placed into separate groups, consisting of no learning (sedentary), physical skill learning and exercising. The exercise group consisted of standard wheel running. In comparison, the physical skill-learning group involved rats having to learn to stay on a rotating rod, with the speed of rotation gradually increasing over time, thus increasing the difficulty of the task as well as creating a need to adjust and relearn. Rats that successfully learned to stay on the rod were found to retain more neurons in their hippocampus than rats from the other groups, even though the exercise group ran twenty times the distance of the learning group. Furthermore, the survival rate of neurons was found to be similar in the exercising and no learning group. The findings indicated that cell survival was related not to the amount of exercise, but to complex skill learning. Based on these findings, Curlik and Shors (2013) suggested that exercises that combine mental and challenging physical skill learning, such as dance, could lead to long lasting effects on the adult brain. Findings with older human adults also show support for the complexity of the physical components of an exercise affecting cognitive outcomes. In humans, coordination exercises have been shown to have a greater positive impact on improving attention and concentration than simple aerobic exercises, possibly due to the complexity of the coordination exercise engaging additional neural networks that are not recruited during simple aerobic exercises (Budde et al., 2008). Such findings may also extend to dance when being compared to other exercises. For example, Coubard et al. (2011) reported that dancing had a significant effect on attentional control in older healthy adults, whereas Tai Chi and fall prevention exercises failed to have any effect. These findings were attributed to the dance intervention consisting of improvised movement, requiring participants to consistently adapt to new motor movements, and thus requiring higher attentional demand, all in contrast to the other two exercises. The inclusion of novel and difficult movements in dance therapy has been suggested to be helpful with improving certain physical impairments found in neurological disorders such as Parkinson's (Duncan and Earhart, 2012). Physical exercise is undoubtedly important, but doing so in a simple and non-stimulating environment may limit its potential in improving cognition (Horne, 2013). Rather, activities such as dancing may be able to provide an enhanced form of exercise due to providing physical novel learning experiences in a stimulating context.

Aside from being a physical activity that includes extensive motor training and novel physical skill learning, dance may also incorporate other elements that can positively influence the impact of a physical activity on neuroplasticity and cognition, such as fostering social interactions. Simply walking with other people has been shown to activate a more widespread brain network compared to walking alone (Saarela and Hari, 2008). Social isolation during running has also been shown to suppress neurogenesis (Stranahan et al., 2006). Yet, social interaction is rarely lacking in dance, due to the two being intrinsically tied to one another. Dancing is most often done with others, either in pairs or groups, with individuals often relying on others for movement cues. However, going even farther back, dance has been speculated to

have originated due to the need for social interaction, acting as a means of non-verbal communication (Brown and Parsons, 2008; Kattenstroth et al., 2013). Ultimately, dance can be seen as possessing various physical activity related properties not found in more conventional exercises, such as motor training and physical skill learning, which can enhance an exercise's impact on neuroplasticity and cognition.

Other potential benefits of dance

Thus far, the potential use of dance for neurorehabilitation has focused on how it simultaneously combines physical and cognitively stimulating activities into one. However, there are other elements of dance that may also contribute to its rehabilitative properties. For example, in regards to the setting of rehabilitation, interventions may be most effective when done in an engaging environment that provides novel and multisensory stimulation (Maegele et al., 2005; Fabel and Kempermann, 2008; Kempermann et al., 2010; Pekna et al., 2012). Animal models support the ability of enriched environments to enhance neuroplasticity through various mechanisms (van Praag et al., 2000), improve cognitive deficits across a variety of neurological disorders (Pang and Hannan, 2013), as well potentially be a more potent therapy for cognitive impairments than physical exercise alone (Will et al., 2004). The very nature of dancing incorporates physical, cognitive and social engagement into a single setting, rich in components that touch upon all these domains; such components include rhythmic motor coordination, balance, memory, emotional engagement, affection, social interaction, acoustic stimulation as well as a musical experience, all of which add up to making dance a human equivalent of an enriched environment (Kattenstroth et al., 2010).

The value of dance as a tool for neurorehabilitation may also extend beyond its potential to improve physical and cognitive impairments. For example, depression is a common comorbid condition associated with various neurological disorders (Rickards, 2005; Hellmann-Regen et al., 2013), capable of not only exacerbating existing symptoms, but also impeding the effects of therapies being undertaken, ultimately reducing quality of life. However, participation in dancing has been shown to be able to significantly improve depression related symptoms (Kiepe et al., 2012; Koch et al., 2014; Vankova et al., 2014). Dancing may also be able, to a certain extent, avoid the adherence issues associated with conventional physical rehabilitation exercises (Dishman, 1988; Van Der Bij et al., 2002; Bassett, 2003). For example, whether an exercise is enjoyable and interesting can influence long term participation (Rhodes et al., 1999; Findorff et al., 2009), something that conventional exercises may not promote. In contrast, dance has been reported as being a highly enjoyable and motivating social activity, leading to adherence in both healthy and clinical groups (Federici et al., 2005; Belardinelli et al., 2008; Westheimer, 2008; Earhart, 2009; Hackney and Earhart, 2009; Heiberger et al., 2011; Houston and McGill, 2013). Factors found in dance that may contribute to this may include it being a social activity; group based exercises have been reported to possess higher participation rates (Van Der Bij et al., 2002), which may in part be explained by the act of developing social relationships being associated with the enjoyment of an exercise (Wankel, 1985). In addition, activities

that are seen as requiring low exertion are favored by older adults (Rhodes et al., 1999). Music also has been noted to enhance participation in exercise, due to lessening the perceived difficulty and discomfort associated with physical activity (Johnson et al., 2001; Schutzer and Graves, 2004).

Thus, dance may be viewed as an interesting alternative therapy for neurorehabilitation due to how it combines various elements into a single experience, including cognitive and physical activities, emotional engagement, social interactions and multisensory stimulation (Kattenstroth et al., 2010), each of which may contribute to its therapeutic value.

CURRENT EVIDENCE FOR THE USE OF DANCE

In regards to investigating the benefits of dance, its use in improving physical functioning has been the focal point. The various physically related benefits that dancing can provide, particularly for older adults, are well known, and includes improving balance, postural control, endurance and motor performance (Hopkins et al., 1990; Shigematsu et al., 2002; Federici et al., 2005; Verghese, 2006; Hui et al., 2009; Keogh et al., 2009; Kattenstroth et al., 2013). These improvements may be attributed to dance being an incredibly comprehensive exercise, including components of both motor and physical fitness, which may allow it to improve flexibility, heighten proprioception, improve muscle strength, and promote balance and posture (Westheimer, 2008). However, a limited number of studies involving both healthy and clinical groups suggest that dancing may also be able to improve cognitive functioning.

In an observational study, Kattenstroth et al. (2010) compared older healthy adults who had on average over a decade's worth of experience in amateur dancing to control matched adults. The amateur dancing group was found to have a superior performance across numerous sensorimotor and cognitive domains in comparison to the no dancing group. However, an important observation was that in relation to the numerous domains that were tested, the difference between the two groups was related to the control group showing poor performances across multiple parameters, and not necessarily the dancing group exhibiting superior performances. This was interpreted as engagement in dance preventing degradation of performance across various cognitive and sensory domains as a result of aging. Indeed, previous findings have suggested that engagement in various lifestyle activities, including dancing, are associated with a lower risk of cognitive decline in older adults (Verghese et al., 2003). However, conflicting results were reported by Verghese (2006), who compared motor and cognitive performances between older social dancers and non-dancers, and found that cognitive test performances did not differ between the two groups. Yet, observational studies do not shed light on the potential use of dance as an intervention tool for improving cognition. Kattenstroth et al. (2013) recently addressed this by investigating the effects of a dancing intervention on older healthy adults. The experimental group consisted of older adults participating in a 6 months dance class, 1 h per week, who were compared to a matched control group. Results showed that participation in a dance class, even for only 1 h per week for 6 months, was able to induce positive effects across various domains. This included significant improvements being found in posture, reaction times,

tactile and motor performances, attention, and across various cognitive domains. Improvements were also greatest in adults who had the poorest performances across the various domains tested before the dance classes took place, suggesting that dance was most effective in those with the greatest impairments prior to the intervention. Unfortunately, there was no long term post-intervention follow up to assess if the improvements reported immediately after the end of the 6 month dance intervention were long lasting. Other studies have also investigated the effects of dance training on cognition in older adults, but have only focused on specific cognitive domains. For instance, Coubard et al. (2011), with an interest on attention in older adults, compared approximately 6 months worth of training in dance classes to Tai Chi and fall prevention programs, and reported that only the dance group showed any improvement on attentional control. Kimura and Hozumi (2012) compared the effects of freestyle and combined dancing on task switching reaction time performance after a single session in healthy older adults, and reported that the combined dancing, by virtue of a decrease in switch cost post-dance class, positively influenced executive functioning, unlike the freestyle dance. However, no control group was included, and the findings, being based on a single session, cannot be used to interpret the impact of long term dance interventions. Thus, although preliminary findings suggest that dancing can influence cognition, there remains insufficient evidence to thoroughly support such a notion.

Dancing interventions have also been used with various neurological populations, aiding with both physical and cognitive impairments. Arguably the most popular application of dance therapy has been with Parkinson's patients, with specific interest on its impact on physical functionality. In regards to aiding with physical impairments, dancing has been noted to include many of the key features recommended in physical therapy for Parkinson's (Keus et al., 2007; Earhart, 2009), including the use of external cues (including through music and movement of partners), teaching of specific movement strategies, incorporation of balance exercises, as well as acting as an aerobic exercise (Earhart, 2009). Indeed, dancing in those with Parkinson's has been found to lead to significant improvements across various physical domains that are commonly impaired, including balance, gait, rigidity, upper extremity functioning and functional mobility (Earhart, 2009; Hackney and Earhart, 2009, 2010; Heiberger et al., 2011; Duncan and Earhart, 2012). Although Parkinson's is primarily described as a movement disorder, it is also associated with impairments in cognition, particularly in executive functioning (Petzinger et al., 2013). However, dance studies involving Parkinson's patients have primarily focused on the potential benefits on physical impairments, and less so on cognition. In a recent study by McKee and Hackney (2013), Parkinson's patients were either enrolled into Tango dance lessons or an educational program, which was designed to include extensive socialization and interactions. In regards to cognition, both groups showed similar improvements in executive functioning following their respective intervention, but only the dance group was found to show any significant improvements in spatial cognition. Gains were also maintained ~10–12 weeks post-intervention. Improvements were suggested to be related to the motor training aspect of dance, since although social interactions may influence the efficacy

of rehabilitation programs, such interactions were believed to be similar between the dance and educational group. Although improvements in executive functioning may in part be explained by dance acting as an aerobic exercise (Colcombe and Kramer, 2003), improvements in spatial cognition were speculated to be related to the dance lessons consisting of structured motor components that required memory of steps and directions, as well as awareness of spatial relationships and patterns in the environment (McKee and Hackney, 2013). Thus, although there is substantial support for the use of dance in aiding with physical and functional impairments in Parkinson's patients, evidence to support its use for improving cognition remains limited.

In regards to the use of dance to improve both physical and cognitive functioning in stroke and dementia patients, research remains sparse. Berrol et al. (1997) examined the effects of a 5 weeks, twice a week, dance class intervention for older stroke and traumatic brain injury patients. The dance intervention was tailored with psychosocial elements in mind, focusing on dance as not only an exercise, but as a creative and social activity. Significant improvements in the dance group were found for balance, self report scores related to social interactions, as well as in cognition, specifically on decision making and short term memory domains. A stroke related case study by Hackney et al. (2012) reported improvements on various physical domains, including balance, mobility and endurance as a result of dancing, although effects on cognition were not investigated. In regards to dementia related populations, Hokkanen et al. (2003) reported in a pilot study that cognition in Alzheimer's patients did not change following a 16 week long dance intervention. Nonetheless, these results do not necessarily mean that dance did not have a positive effect. Rather, this may have implied that participation in dance class prevented the further decline of cognition associated with Alzheimer's. A more recent study by Hokkanen et al. (2008) compared dance therapy in Alzheimer's patients, with the inclusion of both a dance and control group. The dance class was held once a week, for 9 weeks in total. The dance group was found to have improved in specific cognitive domains, including that of visuospatial ability and planning, in comparison to the control group, in which cognitive performances, depending on the domain, remained either stable or declined. Van de Winckel et al. (2004) also reported that dance classes led to significant improvements in cognition in those with dementia. It is important to note that in this study, participation in the dance class was extensive, with sessions being held on a daily basis for 3 months. These findings are in agreement with those of Hokkanen et al. (2008), in the context that dance interventions for older adults with Alzheimer's or dementia may be capable of improving cognition, rather than simply preventing or delaying decline. Similar to findings in healthy and other clinical groups, dancing has also been shown to improve physical functionality, such as strength, balance and gait, in those with Alzheimer's (Abreu and Hartley, 2013).

Undoubtedly, research on dance as a tool for neurorehabilitation is still in its infancy. Yet, with the current evidence limited, particularly for its potential use in aiding with cognition in both healthy and neurological groups, there are still preliminary

findings suggesting that dance can have a positive influence on both cognitive and physical functioning.

CONCLUDING REMARKS

The number of people who will be affected by neurological disorders is expected to increase in the upcoming decades (World Health Organization, 2006). With issues in efficacy of current surgical and pharmacological treatments, as well as conventional rehabilitative therapies, new alternatives are needed. As discussed, physical and musical therapies have been used for aiding with both motor and cognitive impairments across various neurological groups. However, they also have inherent limitations as discussed, creating the need to explore for other alternatives. With its multimodal nature due to simultaneously combining physical and cognitive activity, dance may offer a unique method of rehabilitation that can not only help with physical impairments, but cognitive as well. However, there are some important limitations in regards to the use of dance. Similar to other physical exercises, the recommendation of dance may be limited to those who are physically able; this may exclude patients who are in the most need of aid with physical and cognitive functioning. There may also be an issue with how dance is perceived as a potential treatment. Activities, such as dance, even if prescribed by health professionals, may be seen as recreational and less as a neurorehabilitative therapy. This perception may lead to some not recognizing the clinical value, and ultimately deciding not to participate (Chao et al., 2000).

With the lack of literature on the use of dance for neurorehabilitation, the future undertaking of certain research directions may be imperative to establish its efficacy. Research thus far has shown support for the use of dance in improving various physical domains. However, there is a lack of research on how dance can improve cognition, something that must be addressed if dance is to be promoted as a tool for neurorehabilitation. Future work with dance interventions, in both healthy and neurological groups, may strengthen the literature through the conducting of randomized controlled trials to investigate the affect of dance on cognition. Such interventions could consist of comparing dance classes to other activities, such as physical exercise or musical therapies, to address whether dance holds any unique therapeutic value for physical and cognitive functioning. As suggested earlier, the combination of physical activity with music may lead to an interaction effect, allowing dance to improve cognition in a unique manner compared to physical or musical activities alone; however, studies are needed to substantiate such claims. Investigating the potential impact of dance interventions may also be aided through the use of neuroimaging techniques, which as discussed, have been used to help elucidate how physical exercise and musical activities can enhance neuroplasticity and cognition; as of now, no known studies have been published which investigate changes to the brain as a result of dance interventions. There is also the issue of just what type of dance should be used; usually, the dance classes' style of dance has been dependent on the instructor and their feeling of what the capabilities are of the participants. Dancing comes in various forms and styles, and even though they share similar traits, they also differ in many ways. However, at this point, focus should just be placed on the efficacy of dancing in general. A future line of research may focus

on the efficacy of different types of dances in order to determine which may provide the greatest benefits. Additionally, research may be done with the aim to disentangle the multiple components of dance, such as music, verbal instructions, guided movements learned through visual demonstration, partnered dance either by leading or following, movement through space, and social interactions, in order to get a better sense of which of these may be primarily responsible for the positive changes associated with dance.

Dance, with its multimodal nature, may offer a unique method to address both physical and cognitive impairments in various neurological groups. Its ability to aid with physical functioning has been well documented, and although there is much less research on how dance can positively affect cognition, preliminary findings are nonetheless promising. In comparison to physical and musical therapies, we do not propose that dance undoubtedly holds the greatest impact of the three. Rather, it is most likely that each therapy has its own unique clinical value, as well as limitations. Thus, depending on the circumstances, dance may be a more appropriate and beneficial alternative therapy compared to the other alternatives. Dance may also be able to overcome more practical issues that other therapies may face in their application. This can include it being a low cost therapy that can be done almost anywhere, thanks to little to no equipment needed. It may also, to some extent, avoid adherence issues due to it being a social and enjoyable activity. Thus, dance should be further investigated for its potential use as a tool to not only help with aging gracefully, but more importantly, to help those with their fight against neurological disorders.

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Facing the music: three issues in current research on singing and aphasia

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Keywords: left-hemisphere stroke, speech-language pathology, non-fluent aphasia, apraxia of speech, Melodic Intonation Therapy, singing, rhythmic pacing, formulaic language

Left-hemisphere stroke patients suffering from speech and language disorders are often able to sing entire pieces of text fluently. This finding has inspired a number of music-based rehabilitation programs, most notable among them a treatment known as Melodic Intonation Therapy (Albert et al., 1973). According to the inventors of the treatment, singing should promote a transfer of language function from left frontotemporal neural networks to their preserved right-hemisphere homologues. Although singing indeed engages right frontotemporal areas (Callan et al., 2006; Özdemir et al., 2006), it does not seem to induce a transfer of language function from the left to the right hemisphere (Belin et al., 1996; Jungblut et al., 2014). Nonetheless, several studies confirmed the promising role of singing (Mills, 1904; Gerstmann, 1964; Keith and Aronson, 1975; Tomaino, 2010) and the overall efficacy of Melodic Intonation Therapy (Van der Meulen et al., 2014).

Using an analytic research approach, two recent experiments explored whether singing, rhythmic pacing, and lyric type have an immediate effect on syllable production (Stahl et al., 2011) or a lasting effect on some aspects of speech and language recovery (Stahl et al., 2013). Contrary to earlier reports, the results did not indicate a short- or long-term advantage of singing over rhythmic speech in persons with non-fluent aphasia and apraxia of speech. Rather, the results revealed that lyric type may be of great importance. Conversational speech

formulas—such as “good morning,” “everything alright?” or “I’m fine”—yielded higher rates of correctly produced syllables than novel word sequences, whether they were sung or rhythmically spoken. Moreover, the sung and the spoken training of a few selected speech formulas proved to facilitate the production of these phrases.

We readily acknowledge that increasing the variety of phrases may lead to generalized effects in therapy, while singing in syllable-timed languages such as French possibly adds to the level of rhythmicity in a particular way (cf. Schmidt-Kassow et al., 2011; Zumbansen et al., 2014). Still, this does not fully explain the range of seemingly contradictory findings in the literature. In our opinion paper, we would like to address three issues in current research on singing and aphasia: articulatory tempo, clinical research designs, and formulaic language resources. We believe that these issues may account for some of the major discrepancies between past reports.

ISSUE 1: ARTICULATORY TEMPO

Singing slows down articulatory tempo. This, in turn, has been found to benefit syllable production in patients with speech and language disorders (Beukelman and Yorkston, 1977; Laughlin et al., 1979; Pilon et al., 1998; Hustad et al., 2003). Although the interaction of articulatory tempo and syllable production is often useful in therapy, it may lead to problems in clinical research, as illustrated in a cross-sectional

study (Racette et al., 2006). Eight patients were singing and speaking novel lyrics in two conditions: alone (solo word production), and together with a vocal playback (choral word production). During solo word production, the number of intelligible words was independent of whether the patients were singing or speaking. During choral word production, the number of intelligible words was generally higher, with more sung than spoken words articulated correctly. Based on these results, one may conclude that choral singing facilitates word production in aphasic patients.

Taking a closer look at the experimental design, however, some details of the study are worth noting. The sung playback voice produced words only half as fast as the spoken playback voice. This difference also affected the patients’ actual performance. During solo word production, the patients were relatively free to choose their preferred articulatory tempo (mean [M] duration of sung syllables: $M = 572$ ms; spoken syllables: $M = 494$ ms; $\Delta = 78$ ms; cf. Racette et al., 2006). During choral word production, the patients adapted to the articulatory tempo of the sung and spoken vocal playbacks (sung syllables: $M = 696$ ms; spoken syllables: $M = 426$ ms; $\Delta = 270$ ms). Hence, the mean difference in syllable duration between singing and speaking (Δ) was 3.5 times larger during choral word production than during solo word production.

This comparison reveals that the patients had more time to articulate well when they were singing to vocal

playback. Without a doubt, the reported results suggest a facilitating effect of choral word production, whether sung or spoken. However, the results do not necessarily indicate a benefit from choral singing. What seems like a choral singing effect may actually arise from differences in articulatory tempo. Using a similar research design, but controlling for articulatory tempo, a later study did not confirm an effect of choral singing over choral speech in 17 patients (Stahl et al., 2011). The consistent duration of sung and spoken syllables may be one of the reasons for this contrary finding. In summary, the control of articulatory tempo appears to be crucial in studies that seek to determine how different forms of vocal expression affect syllable production in patients with speech and language disorders.

ISSUE 2: CLINICAL RESEARCH DESIGNS

Another issue concerns clinical studies that address the underlying mechanisms of music-based aphasia therapy. One seminal treatment study investigated the efficacy of Melodic Intonation Therapy (Schlaug et al., 2008). A language test indicated that Melodic Intonation Therapy was more effective than a non-melodic control treatment in 2 chronic aphasic patients. Although the results may not generalize to a larger clinical population, they nonetheless provide evidence for the efficacy of Melodic Intonation Therapy in a group of chronic aphasic patients. However, it is worth considering whether or not the results offer insight into the efficacy of any particular therapeutic element included in the program—such as singing—and its underlying neural mechanisms, as revealed by structural and functional imaging.

It is important to note that Melodic Intonation Therapy includes various forms of vocal expression and multi-modal feedback: singing minor thirds; rhythmic speech with exaggerated prosody (*sprechgesang*); tactile rhythmic stimulation via hand tapping; choral word and phrase production; solo word and phrase repetition; auditory cueing of initial word and phrase syllables; and so on (cf. Helm-Estabrooks et al., 1989). Given the number of stimulating elements used in Melodic Intonation Therapy, it is challenging to

assess the contribution of each element to the efficacy of the entire program. This problem becomes especially apparent if one compares the composition of non-melodic control treatments and Melodic Intonation Therapy in clinical studies.

In the study mentioned above, the non-melodic control treatment did not include singing, rhythmic *sprechgesang*, and rhythmic hand tapping, whereas Melodic Intonation Therapy did (cf. Schlaug et al., 2008). That is, the treatments did not only differ in singing, but also in other aspects of vocal expression and sensorimotor feedback. Consequently, the results do not necessarily support the clinical efficacy of singing and its underlying neural mechanisms. What seems like a benefit from singing may actually be the effect of rhythm, prosody, tactile stimulation, or any of their combinations. Considering this range of possible interpretations, it is not a contradiction that a recent experiment did not confirm a long-term effect of singing over rhythmic speech in 10 chronic aphasic patients (Stahl et al., 2013). In summary, the interpretation of clinical results depends on whether the underlying research design focuses on music-based aphasia therapy as an entire program or on its specific mechanisms.

ISSUE 3: FORMULAIC LANGUAGE RESOURCES

One reason for the success of music-based aphasia therapy may be its use of common phrases. The original manual of Melodic Intonation Therapy proposes phrases such as “I love you,” “how are you?” or “thank you” at the lower proficiency level of the program (Helm-Estabrooks et al., 1989). The phrases are stereotyped in form, tied to social context and, therefore, fall into the category of formulaic language (Van Lancker Sidtis, 2004). This fact is critical: according to present knowledge, the production of formulaic language engages bilateral neural networks including right frontotemporal areas, the right basal ganglia and, possibly, the right cerebellum (Hughlings-Jackson, 1878; Speedie et al., 1993; Ackermann et al., 1998; Van Lancker Sidtis et al., 2003; Sidtis et al., 2009). The degree of right-hemisphere lateralization seems to be strongest for pragmatically oriented formulaic language: pause fillers, discourse elements, and conversational

speech formulas (Van Lancker Sidtis and Postman, 2006). It is intriguing to consider what these results may imply for clinical practice.

Formulaic expressions may be viewed as a valuable language resource in patients with left-hemisphere lesions. Recent evidence suggests that standard speech-language therapy facilitates newly created utterances, while the massive repetition of conversational speech formulas leads to long-term progress in the production of trained phrases (Stahl et al., 2013). Eight out of ten patients were able to establish their own individual formulaic repertoire to communicate some basic needs in daily life. Moreover, the therapeutic use of conversational speech formulas may have a motivating and rewarding effect, most notably in patients with extended left frontotemporal and subcortical lesions. Patients often report feeling competent if they are able to produce a few phrases correctly. Finally, it is conceivable that even non-formulaic expressions become part of the formulaic repertoire by means of massive repetition: patients may eventually retrieve the trained phrases as a coherent unit from the mental lexicon, engaging bilateral or right-hemisphere neural networks (cf. Wolf et al., 2014).

The crucial role of formulaic language in music-based aphasia therapy challenges the interpretation of structural and functional neuroimaging data. Until recently, the sensitivity of right frontotemporal areas to Melodic Intonation Therapy has been presumed to result from the neural plasticity of non-formulaic language (Schlaug et al., 2008, 2009; Vines et al., 2011). However, the imaging data may also depend on the plasticity of right-hemisphere neural networks that support the production of formulaic language. What seems like a melody-mediated transfer of language function from the left to the right hemisphere may actually be the use of right-hemisphere language resources. Behavioral evidence for this claim comes from the finding that music-based aphasia therapy benefits the production of conversational speech formulas, irrespective of whether the patients are singing (Stahl et al., 2013). In summary, future research needs to consider the possible interplay of music-based aphasia therapy and right-hemisphere neural networks

engaged in the production of formulaic language.

OUTLOOK ON FUTURE RESEARCH

There is no doubt that music-based aphasia therapy—including Melodic Intonation Therapy—is a promising treatment for several types of speech and language disorders. The rhythmic elements of the program may help to overcome deficits in motor planning, commonly found in persons with apraxia of speech. The training of formulaic and non-formulaic phrases may indirectly compensate for communicative difficulties associated with agrammatism, as is the case in persons with Broca's aphasia. The repetitive character of the program as well as the focus on a limited formulaic repertoire may be suitable to alleviate severe expressive and receptive symptoms, typically observed in persons with global aphasia. In other words, the use of melody-based aphasia therapy seems appropriate for a number of speech and language disorders.

To this day, research on music-based aphasia therapy has addressed the overall efficacy of current rehabilitation programs (holistic approach) and some of their underlying mechanisms (analytic approach). Drawing on the findings from holistic approaches, the long-term goal of analytic research is to tailor future rehabilitation programs to the individual needs of the patients. This may increase the efficacy of the treatment to a considerable extent. One may argue that analytic research on music-based aphasia therapy is taking a reductionist view, for example, by disentangling the close relationship between melody and rhythm (cf. Merrett et al., 2014). Until now, however, analytic research on music-based aphasia therapy has actually compared singing—that is, the *combined* use of melody and rhythm—with other forms of vocal expression, including rhythmic speech. The available data are, therefore, consistent with the idea that rhythmicity is naturally inherent in singing.

Still, analytic research on music-based aphasia therapy should not overshadow the valuable contribution of holistic approaches. We believe that both holistic and analytic approaches are needed and usually depend on each other. Many

clinical hypotheses are derived from analytic research and then tested in holistic designs, and vice versa. Acting in concert, holistic and analytic approaches may help to improve the quality of research in the field as well as the individual efficacy of music-based aphasia therapy.

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Intracerebral functional connectivity-guided neurofeedback as a putative rehabilitative intervention for ameliorating auditory-related dysfunctions

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Electroencephalography (EEG) constitutes one of the most eligible candidates for neurofeedback applications, principally due to its excellent temporal resolution best reflecting the natural dynamics of brain processes. In addition, EEG is easy to use and provides the opportunity for mobile applications. In the present opinion article, we pinpoint the advantages of using intracerebral functional connectivity (IFC) instead of quantitative scalp EEG for interventional applications. In fact, due to the convergence of multiple signals originating from different spatial locations and electrophysiological interactions, miscellaneous scalp signals are too unspecific for therapeutic neurofeedback applications. Otherwise, IFC opens novel perspectives for influencing brain activity in specific dysfunctional small- and large-scale neuronal networks with a reasonable spatial resolution. In the present article, we propose concrete interventional IFC applications that may be used to ameliorate auditory-related dysfunctions such as developmental dyslexia.

Keywords: functional connectivity, EEG, neurofeedback, developmental dyslexia, rehabilitation, auditory-related cortex

FROM HISTORICAL BRAIN PERSPECTIVES TO MODERN NEUROSCIENTIFIC APPROACHES

During the 19th century, brain researchers took advantage of individuals suffering from brain lesions in order to determine the contribution of specific brain areas to different aspects of behavior, including perception, speech processing, motor skills, and cognitive functions (Zolamorgan, 1995; Leff, 2004). During the same century, an intellectual quarrel raged between researchers who believed that brain functions are localized in separable brain areas (localization view) and those who argued that the entire or parts of the cortex contributes to behavior (network view; for an historical overview see for example Leff, 2004). Nowadays, one can draw some hazardous analogies between modern neuroscientific approaches and historical perspectives on brain functions depending on the imaging technique used. In fact, functional magnetic resonance imaging (fMRI) has a very poor temporal resolution, leading to the illusory impression that specific brain functions are localized in distinct brain areas. Otherwise, due to the dynamic and blurred nature of electrical scalp signals (i.e., electroencephalography, EEG), one could naively come to the conclusion that the entire brain contributes to a specific behavior.

Currently, it is generally acknowledged that widely distributed, specialized, and dynamic cortical-subcortical networks form the fundamental basis of behavior (Bullmore and Sporns, 2009). Within this framework, EEG has gained more and more attention in the field of cognitive neuroscience, mainly due to its excellent temporal resolution (in the range of milliseconds) enabling to

capture the dynamic dimension of brain functioning in a more realistic manner than neuroimaging does. In addition, based on novel mathematical applications it is now possible to estimate the intracerebral origin of scalp signals (Scherg, 1990; Pascual-Marqui et al., 1994) as well as to objectify intracerebral functional connectivity (IFC) in real-time (Canuet et al., 2011; Kühnis et al., 2014) with a reasonable spatial resolution (Pascual-Marqui et al., 1994; Phillips et al., 2002). Thus, EEG is particularly suitable for comprehending the dynamic interplay between specific brain regions within local and global neuronal networks in both natural and dysfunctional brain conditions.

NEURONAL NETWORKS: THE BEARING SKELETON OF BRAIN FUNCTIONS

Currently, there is no doubt that cognition (Langer et al., 2013), motor functions (Jin et al., 2012), and perception (Wu et al., 2012) do not function in isolation but are embedded in neuronal assemblies consisting of networks influencing each other's through excitatory and inhibitory signals (Fell and Axmacher, 2011). Such small- and large-scale neuronal networks can be represented by using both functional and structural data as well as by taking into account different parameters, like white matter integrity (Hagmann et al., 2007; Elmer et al., accepted), cortical thickness (Hanggi et al., 2011), cortical surface area or volume (Bonilha et al., 2004; Sanabria-Diaz et al., 2010), hemodynamic responses (Rehme et al., 2013), or even intracerebral oscillatory phase synchronization values (Langer et al., 2012; Kühnis et al.,

2014). Such neuronal networks can for example be modeled by taking into account mathematical graph theories (i.e., small-world networks) where most nodes within a network can be reached from every other by a small number of steps. This implies that efficient systems with small-world topology are characterized by a high local clustering coefficient (i.e., the degree to which nodes in a graph tend to cluster together) and short path lengths between distant nodes (Bullmore and Sporns, 2009). The advantage of focusing on such networks rather than on localized brain characteristics is that the former can support both segregated/specialized as well as distributed/integrated information processing (Bullmore and Sporns, 2009).

From a functional perspective, it is assumed that brain regions that do the same at the same time are somehow interconnected (i.e., functional connectivity). Functional connectivity and systemic brain organization can be described by using dynamic causal modeling (Eickhoff et al., 2009), Granger causality (Jancke, 2012), or correlative analyses between brain signals (for example signal amplitude, current density, power, or phase synchronization) originating from different spatial locations (Langer et al., 2012). Even though it results evident that structural and functional brain properties are mutually related (Miranda-Dominguez et al., 2014), the advantage of focussing on functional connectivity is that it enables to capture the dynamic nature of the human brain in different time-scales, ranging from milliseconds (i.e., EEG) to several seconds (i.e., fMRI).

EEG AND INTRACEREBRAL FUNCTIONAL CONNECTIVITY

The discovery of EEG by Berger (1929) can be considered as one of the most important historical breakthrough in the field of neurology and cognitive neuroscience. This specific technique builds up on single electrodes that are fixed on the surface of the scalp for recording electrical brain activity. Through different applications in the field of electrical engineering (i.e., signal amplification, impedance reduction, etc.), it became possible to measure the summed electrical postsynaptic activity that is locked or unlocked to an external (for example auditory stimulation), or internal (for example imagery) event at the surface of the scalp. Such electrical brain activity can be quantified, for example, by evaluating the amplitude and timing of event-related potentials (ERPs), power spectra in different frequency ranges over time, or the degree of phase alignment (i.e., coherence) in a specific frequency band between single scalp electrodes.

In the last 15 years, novel mathematical applications render it possible to overcome the so called “inverse problem” of intracerebral EEG source estimation (Pascual-Marqui et al., 1994) however, with some drawbacks in terms of spatial resolutions (Phillips et al., 2002). Currently, several toolboxes and software (http://en.wikipedia.org/wiki/Comparison_of_neurofeedback_software) can be used for estimating intracerebral brain activity based on the electrical signal recorder from the surface of the scalp. These new technologies imply that for each signal measured on the surface of the scalp it becomes possible to estimate intracerebral brain activity for each voxel, Brodmann area, or region of interest (ROI) in the form of current-, or spectral-power density by retaining phase information. Therefore, all these measures can be taken for modeling IFC networks (see previous section).

In turn, we will provide two examples of practical applications of IFC in a specific group of experts, namely professional musicians. In a first study, we measured professional musicians and non-musicians by using EEG and IFC analyses. We postulated that auditory-specialization (Elmer et al., 2012; Marie et al., 2012; Kuhnle et al., 2013) and asymmetry (Schneider et al., 2002) in musicians should be dependent, at least in part, by the amount of interhemispheric communication between the left and right auditory-related cortex (ARC). Based on this assumption, we measured intracerebral phase synchronization (see **Figure 1**) in the theta, alpha, and beta frequency range between the two ARC in musicians and non-musicians. We found support for our hypothesis in that musicians showed increased IFC between the two ARC as well as a relationship between IFC and the amplitude of auditory-evoked potentials (Kühnis et al., 2014).

A second example that depicts a relationship between IFC and expertise arises from a recent study of our group (Elmer et al., under revision) where we tried to integrate two apparently opposite perspectives on absolute pitch, that is the ability to recognize the chroma (i.e., pitch) of a tone without a reference tone (Levitin and Rogers, 2005). In this context, some researchers argue that this specific ability relies on an optimized “early categorical perception” at the processing level of the left ARC (i.e., perception; Siegel, 1974), whereas others suggest that the distinctive trait of AP more likely derives from mnemonic facilitation (Elmer et al., 2013) enabling “pitch labeling” mechanisms by recruiting left-sided prefrontal brain regions (i.e., cognition; Zatorre et al., 1998). By combining EEG and resting-state IFC, we evaluated phase synchronization between the left ARC and the left dorso-lateral prefrontal cortex in the theta ($\sim 4\text{--}7$ Hz) frequency range in a group of musicians with and without AP. Theta oscillations have previously been shown to reliably reflect mnemonic processes (Kahana et al., 1999; Caplan et al., 2001; Ward, 2003; Sauseng et al., 2005), information integration (Ward, 2003), and neuronal communication between distinct brain regions over long-range circuits (Ward, 2003; Polania et al., 2012). Results revealed that in AP musicians perceptual and cognitive subdivisions of the human brain

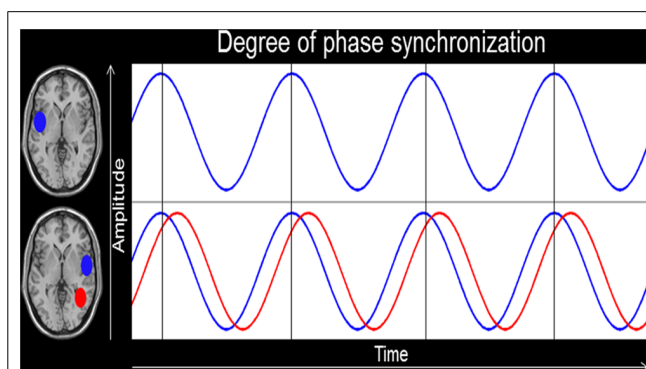


FIGURE 1 | Intracerebral functional connectivity (IFC). This figure depicts the degree of phase alignment between two regions of interests (ROIs) in the left and right hemisphere. The bottom blue oscillation provides an example of perfect phase synchronization, whereas the red one shows a temporal lag in phase synchronicity.

are tightly coupled through oscillatory theta phase-alignment. In addition, within the AP group this specific electrophysiological marker was predictive of pitch-labeling performance by explaining about 30% of behavioral variance. These two EEG studies target at illustrating practical applications of IFC analyses for evaluating systemic brain reorganizations rather than focusing on localized brain functions in isolation. This point of view is also supported by a recent paper of Seither-Preisler et al. (2014) providing specific evidence for increased bilateral synchrony of the primary auditory evoked responses collected at the surface of the scalp in children undergoing musical training compared to children suffering from attention-deficit hyperactivity disorder. Interestingly, this functional dysalignment of auditory-evoked brain responses was accompanied by anatomical specificities of auditory-related brain regions.

THE BASIC PRINCIPLES OF INTRACEREBRAL FUNCTIONAL CONNECTIVITY-GUIDED NEUROFEEDBACK

Neurofeedback bases on cybernetic models consisting of using information about the physiological state of an organism for changing it in a specific direction (Gunkelman and Johnstone, 2005). Such cybernetic models can be utilized when the system to be analyzed is assumed to rely on closed signal-loops. This means that a change in a biological system (in this case brain activity) generates specific changes in the environment (in our case the feedback) that on his part triggers a modulation of the biological system (i.e., brain activity; see **Figure 2**). Due to the high temporal resolution of EEG as well as to novel mathematical applications, it is now possible to modulate the own brain activity in quasi

real-time based on a specific feedback (i.e., visual, auditory, haptic, etc.). Meanwhile, there is a vast body of literature describing neurofeedback applications in several fields of clinical neuroscience (Schoenberg and David, 2014), including the treatment of addiction (Dehghani-Arani et al., 2013), attention-deficit hyperactivity disorder (Maurizio et al., 2014), depression (Young et al., 2014), epilepsy (Tan et al., 2009), and much more (Schoenberg and David, 2014).

An important prerequisite for clinical neurofeedback applications is to exactly know which intra- and extracerebral EEG parameters best reflect a specific natural or dysfunctional brain condition. In addition, an accurate identification of dysfunctional brain areas as well as of functional networks constitutes an important step toward evidence-based clinical applications. In the present article, we principally focus on IFC-guided neurofeedback rather than on the modulation of brain signals at the surface of the scalp. This line of argumentation is supported by the fact that scalp-signals are composed of miscellaneous and unspecific brain activity originating from a variety of brain regions. Furthermore, we are of the opinion that it is more efficient to dynamically change IFC between specific brain regions of interest instead of focusing on the modulation of restricted brain functions, than the former approach more likely takes into account the dynamic and interconnected nature of the human brain (see previous sections).

IFC guided neurofeedback applications base on exactly the same cybernetic models described in the previous section and depicted in **Figure 2**. However, in contrast to scalp-data based neurofeedback, it is necessary to postulate clear assumptions on specific brain areas that are dysfunctional as well as on functional connectivity between these areas. Therefore, the first step of IFC-based neurofeedback is the identification of ROIs within a dysfunctional network. In addition, depending on the connectivity parameters to be trained (i.e., current density, phase synchronization, etc.) one should have a clear conception of the direction of modulations, that means increased or reduced intracerebral activity within the network of interest. In the case of IFC-based neurofeedback relying on the modulation of oscillatory phase alignment between different brain regions, specific knowledge about the relationships between brain functions and oscillations (i.e., delta, theta, alpha, beta, or gamma) is strictly required. In turn, we will describe the pathogenesis of a specific neurological disorder that is associated (at least in part) with auditory-related dysfunctions, namely developmental dyslexia. Based on a review of current research literature, we will propose concrete IFC-based neurofeedback applications relying on the entrainment of lagged phased synchronization. The modulation of lagged phase synchronization constitutes a fruitful approach in that this measure is supposed to reflect true connectivity by taking into account the delay of impulse propagation that is influenced by volume conduction (Pascual-Marqui et al., 2011). It is important to mention, that the ARC shows a huge inter-individual variability (Steinmetz et al., 1989; Zatorre, 2013) and asymmetry (Marie et al., 2013), and that this variability is additionally strongly influenced by training and expertise (Schlaug et al., 1995). Therefore, depending on the research question addressed and on the sample of subjects studied, it is important to take into account such influencing variables.

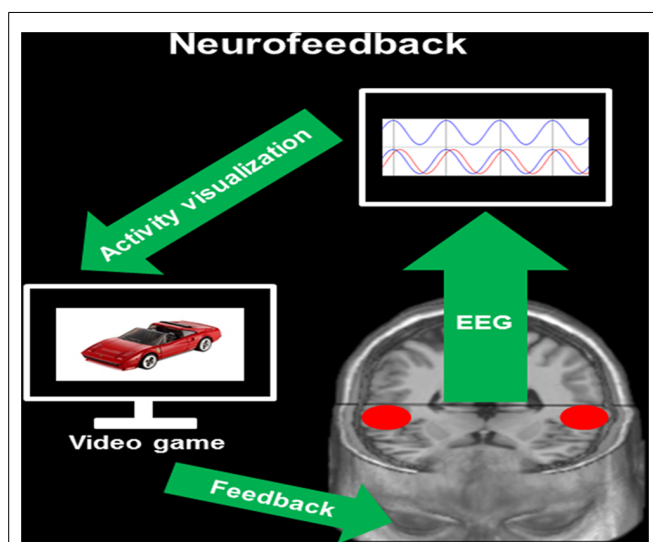


FIGURE 2 | Neurofeedback. This figure provides a simplified overview of IFC-based neurofeedback. During EEG recording, the neurofeedback software provides information about the degree of phase alignment in a *priori* defined intracerebral ROIs [here the bilateral auditory-related cortex (ARC), red circles]. This information is visualized on a monitor by means of a brain-computer interface (activity visualization). Through the coupling of brain activity with a specific task (here video game), participants receive a visual feedback on the modulation of the own brain activity (feedback, here lagged phase synchronization).

DEVELOPMENTAL DYSLEXIA

The main purpose of the present work is to discuss concrete applications of IFC-based neurofeedback for the treatment of auditory-related dysfunctions. However, due to profound differences in the pathogenesis of such dysfunctions, here we will focus on developmental dyslexia only. It is important to mention that it is conceivable that similar approaches we present in association with dyslexia can be extended to other auditory-related dysfunctions, like for example tinnitus (Okamoto et al., 2010) or developmental language disorders (Heim et al., 2013).

Developmental dyslexia can be described as low reading and writing skills despite average intelligence, good educational support, and solid social background (Habib, 2000; Demonet et al., 2004). In the last three decades, several theories have been proposed for explaining the specific deficits in developmental dyslexia, including general perceptual/phonetic- (Tallal and Piercy, 1973; Merzenich et al., 1996; Stein, 2001; Goswami et al., 2002), attentional- (Bogon et al., 2014), and working memory deficits (Ahissar et al., 2006). Also visual (Lovegrove et al., 1980; Stein, 2012) and motor impairments (Nicolson and Fawcett, 1990) have been described.

Several of these theories postulate that developmental dyslexia is somehow related to auditory-related dysfunctions (Tallal and Piercy, 1973; Goswami et al., 2002). The “rapid processing deficit theory” proposed by Tallal and Piercy (1973) and Merzenich et al. (1996) postulates a specific impairment in the processing of fast-changing verbal cues, such as formant transitions and voice-onset time (VOT). In a similar way, Goswami et al. (2002) postulated that dyslexia is associated with a poor temporal resolution of speech sounds that specifically affects the processing of sound rise time. Other theories on dyslexia are rather centered on phonological abilities (Stanovich, 1988; Serniclaes et al., 2001; Ramus, 2003; Ramus et al., 2013) and base on the assumption that dyslexic individuals are specifically impaired in building-up phonological representations (Stanovich, 1988). Finally, also impaired phonological awareness (Ramus, 2003; Ramus et al., 2013) and abnormal sensitivity to within phonemic category variations (Serniclaes et al., 2001) have previously been proposed to constitute the salient trait of developmental dyslexia. For a more comprehensive review of the literature on dyslexia, the reader is addressed to a previous work of Hamalainen et al. (2013).

Interestingly, most of the theories described above are compatible, at least in part, with the view that dyslexic children often show functional (Blau et al., 2009, 2010; Kast et al., 2011) and structural (Hugdahl et al., 2003; Brambati et al., 2004; Bloom et al., 2013) variations in the left ARC, a brain region that is relatively strongly involved in the processing of fast changing verbal and non-verbal cues and phonemes (Zatorre and Belin, 2001; Griffiths and Warren, 2002; Hickok and Poeppel, 2007). In addition, previous fMRI (Zatorre and Belin, 2001; Griffiths and Warren, 2002; Shaywitz et al., 2003, 2007; Hickok and Poeppel, 2007), DTI (Hoeft et al., 2011), and EEG (Dujardin et al., 2011) studies provided evidence for a stronger recruitment of right-sided ARC in dyslexic individuals during speech processing, possibly for compensating poor left-sided temporal resolution.

In a recent multi-pattern neuroimaging study Boets et al. (2013) reported intact phonetic representations (in terms of robustness

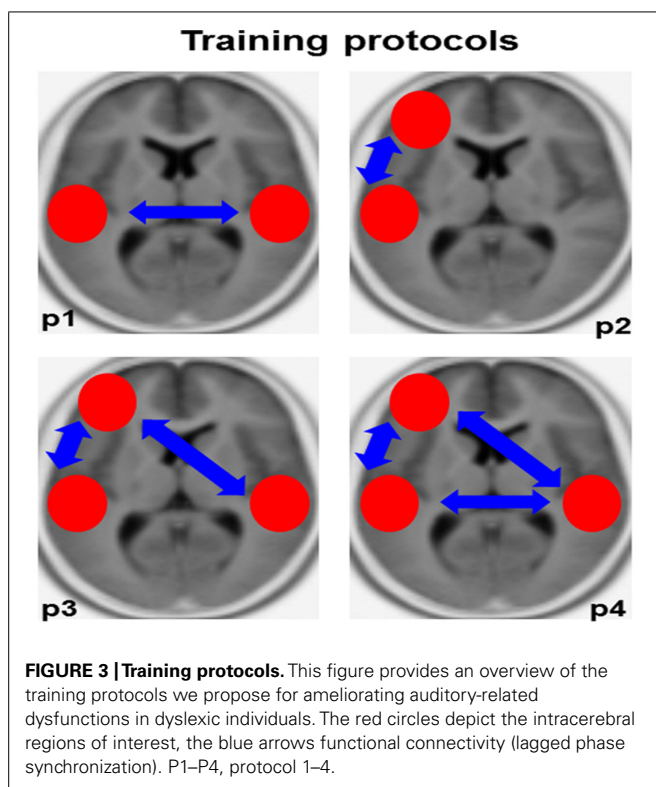
and distinctness) in the bilateral ARC in adults suffering from dyslexia. Most notably, by combining functional and structural connectivity analyses, the same authors’ revealed reduced connectivity between bilateral auditory-related brain regions as well as between the auditory cortices and the left inferior frontal gyrus, the latter region being involved in higher order cognitive functions. These results are interesting in that they open the possibility to consider dyslexia as a neuropsychological state where not phoneme representation *per se*, but rather the access to these representations, is dysfunctional. A similar perspective can be taken into account when considering a recent publication of Vandermosten et al. (2013) where the authors combined DTI and EEG measurements in a sample of adult dyslexic individuals and found evidence for reduced white matter lateralization in the left posterior supratemporal plane and arcuate fasciculus. In addition, white matter lateralization in the posterior superior temporal gyrus and white matter integrity in the posterior part of the corpus callosum were related to phase coherence in bilateral auditory-related brain regions in the frequency range roughly corresponding to phonemic-rate modulations (~ 20 Hz, β). Meanwhile, there is even evidence from longitudinal studies (Langer et al., 2013) showing that functional connectivity can change after only few weeks of training.

INTRACEREBRAL CONNECTIVITY-GUIDED NEUROFEEDBACK AS A PUTATIVE REHABILITATIVE INTERVENTION FOR DEVELOPMENTAL DYSLEXIA

As described in the previous section, there is strong evidence showing dysfunctional left-sided (Rumsey et al., 1992; Temple, 2002) and compensatory right-sided (Shaywitz et al., 2003, 2007; Dujardin et al., 2011) brain activity in the ARC of dyslexic individuals. In addition, recent data point to altered functional (Poelmans et al., 2012; Vandermosten et al., 2013) and structural (Boets et al., 2013) connectivity among bilateral auditory-related brain regions as well as between the bilateral ARC and the left inferior frontal gyrus (Boets et al., 2013). The latter brain region is supposed to be involved in accessing higher-order phonological representations. With these previous results in mind, we will propose specific IFC-guided neurofeedback protocols that may be useful for ameliorating the auditory-related impairments often observed in dyslexic individuals. Please consider that these neurofeedback protocols are ordered in a hierarchical manner that means from small- to large-scale network reorganization.

TRAINING PROTOCOL 1

Based on the often observed hypoactivity of the left ARC in conjunction with the compensatory hyperactivity of its right-sided homolog in dyslexic individuals, we propose a training protocol targeting at ameliorating the division of labor (i.e., intracerebral lagged phase synchronization) between these two perisylvian brain regions (i.e., Brodmann areas 41/42/22). The reasoning beyond this training protocol is that the amelioration of functional connectivity between bilateral auditory-related brain regions may improve the functional capacity of the left ARC and at the same time reduce right-sided compensatory activity (Figure 3, p1). Along this vein, it is conceivable that an increase in phase alignment in at least two frequency bands may possibly improve reading



skills, namely theta ($\sim 4\text{--}7$ Hz) and beta ($\sim 13\text{--}20$ Hz). In fact, theta oscillations roughly overlap with the processing rate of syllables, whereas beta oscillations coincide with the temporal dynamics of single phonemes (Poeppel, 2003; Poelmans et al., 2012). This reasoning is in line with previous work showing that dyslexic individuals are characterized by poor phonological awareness (Ramus, 2003; Ramus et al., 2013), a condition that is strongly dependent on the segmentation of single words into smaller units, namely syllables and phonemes.

Finally, it is important to mention that two previous studies of our group highlighted a relationship between the superiority of professional musicians (compared to non-musicians) in processing segmental speech cues (i.e., syllables varying in VOT and vowels) and functional (Kühnis et al., 2014) as well as structural (Elmer et al., accepted) connectivity among bilateral auditory-related brain regions. Based on these previous results, we believe that the improved and optimized auditory system of musicians can provide fruitful information for developing novel rehabilitative neurofeedback strategies targeting at optimizing auditory-related dysfunctions in dyslexic individuals. Certainly, future studies focusing on the validation of the training protocols we present in the present work are strictly required for optimizing rehabilitative power.

TRAINING PROTOCOL 2

Previous work has described reduced functional and structural connectivity between the left ARC and the left inferior frontal gyrus in dyslexic individuals (Boets et al., 2013). Therefore, we may speculate whether in dyslexic individuals (probably mainly adults) not phoneme representation *per se*, but rather the access to

these mnemonic representations in the left inferior frontal gyrus, is dysfunctional (Boets et al., 2013; Vandermosten et al., 2013). Based on previous work showing that neuronal oscillations in the theta-frequency range ($\sim 4\text{--}7$ Hz) reflect mnemonic processes (Kahana et al., 1999; Caplan et al., 2001; Ward, 2003; Sauseng et al., 2005; Elmer et al., under revision), information integration (Ward, 2003), and neuronal communication between distinct brain regions over long-range circuits (Ward, 2003; Polania et al., 2012), we propose the possibility to ameliorate the recruitment of higher order phonetic representations by increasing theta phase synchronization between the left ARC (BA 41/42/22) and the left inferior frontal gyrus (BA 44/45/47). See Figure 3, p2.

TRAINING PROTOCOL 3

The third training protocol we propose here is an extension of “training protocol 2” (Figure 3, p3). Subjects are trained to increase intracerebral phase synchronization in the theta frequency range simultaneously between both the left and right ARC and the left inferior frontal gyrus (Boets et al., 2013).

TRAINING PROTOCOL 4

This protocol implies a simultaneous combination of training protocols 1 and 3 (Figure 3, p4).

FUTURE PERSPECTIVES

In the present opinion paper we discussed the possibility to ameliorate auditory-related dysfunctions by using IFC-based neurofeedback application targeting at changing the systemic functional brain organization rather than focusing on brain functions in isolation. It is important to mention that here we only addressed some putative application without any claim to completeness. In addition, we want to emphasize that future studies are strictly required for evaluating the rehabilitative relevance of the single training protocols we propose. We explicitly abstained from providing indications on specific training parameters (i.e., training duration and frequency) because we are of the opinion that neurofeedback therapists are best skilled for arranging and optimizing the training protocols we propose. Finally, it is important to remark that in our opinion a better understanding of simple connectivity circuits should be the first step. Only after having collected enough evidence for valid therapeutic applications in small-brain circuits, it makes sense to consider more systemic brain reorganization.

CONNECTIVITY TOOLBOXES

For an overview of different neurofeedback applications, the reader is addressed to the following Wikipedia page: http://en.wikipedia.org/wiki/Comparison_of_neurofeedback_software

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Basics for sensorimotor information processing: some implications for learning

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In sensorimotor activities, learning requires efficient information processing, whether in car driving, sport activities or human–machine interactions. Several factors may affect the efficiency of such processing: they may be extrinsic (i.e., task-related) or intrinsic (i.e., subjects-related). The effects of these factors are intimately related to the structure of human information processing. In the present article we will focus on some of them, which are poorly taken into account, even when minimizing errors or their consequences is an essential issue at stake. Among the extrinsic factors, we will discuss, first, the effects of the quantity and quality of information, secondly, the effects of instruction and thirdly motor program learning. Among the intrinsic factors, we will discuss first the influence of prior information, secondly how individual strategies affect performance and, thirdly, we will stress the fact that although the human brain is not structured to function errorless (which is not new) humans are able to detect their errors very quickly and (in most of the cases), fast enough to correct them before they result in an overt failure. Extrinsic and intrinsic factors are important to take into account for learning because (1) they strongly affect performance, either in terms of speed or accuracy, which facilitates or impairs learning, (2) the effect of certain extrinsic factors may be strongly modified by learning and (3) certain intrinsic factors might be exploited for learning strategies.

Keywords: sensorimotor activities, reaction time, errors, learning, information processing

Whether in sport, car driving, music, or human–machine interactions, learning sensorimotor activities requires efficient information processing. Knowing some principles of human information processing may be useful to propose tasks and/or learning methods in conformity with these principles, so as to facilitate their realization and improve performance. Presenting some basics of these principles is the aim of the present article.

Even when the level of vigilance is optimal, the efficiency of such processing can be affected by extrinsic (task-related) or intrinsic (subject-related) factors. We call here “extrinsic” those factors on which the subject cannot act. Extrinsic factors may be related to the quantity of stimuli to be processed, the way decisions must be taken, the ability to exploit and consolidate appropriately motor programs or the availability of prior information regarding future events. We call here “intrinsic” those factors on which the subject can act. They may be related to strategic effects, voluntary orientation of attention (*in time and space*), motor preparation (*in time and space*) or action monitoring. In fact, intrinsic factors could be unified in terms of executive control processes (Norman and Shallice, 2000).

We will, first, discuss of extrinsic factors and, secondly, present intrinsic ones with a special emphasis on action monitoring processes.

EXTRINSIC FACTORS

STIMULUS-RELATED

We will not discuss here the effects of the quality of the stimuli since it is a commonplace that degraded or ambiguous stimuli

impair perception and identification processes, which results in impaired performance.

Number of relevant stimuli

The number of stimuli to be processed is a critical determinant of performance.

If the stimuli are presented sequentially, increasing their number by time unit amounts increasing the time pressure put on the task, which will result in an increase of the error rate, according to the well established speed–accuracy trade-off (SAT; Fitts, 1966; Pew, 1969; Usher et al., 2002; Bogacz et al., 2009). We will briefly present SAT in section 2 (intrinsic factors).

In case several relevant stimuli must be presented simultaneously, increasing their number will force subjects to share their attention between these different stimuli, which will increase the reaction time (RT) and the likelihood to produce errors. This increase is explained by the fact that, all other things being equal, (1) processing of a relevant stimulus will interfere with processing of other relevant ones, because human information processing capacities are limited (Kahneman, 1973; Norman and Bobrow, 1975; Franconeri et al., 2013); (2) the mere fact to have one’s attention to be shared between several stimuli may often induces a cost, *per se* (Navon and Gopher, 1979). This cost may also be manifested by an increase in RT and/or an increase in error rate. Therefore, if performance is critical, attention sharing, when possible, should be avoided in sensorimotor activities (Hyman, 1953). We do not intend to discuss this point further since accurate and comprehensive

reviews on this subject are already available (e.g., Wickens, 2002, 2008).

Number of irrelevant stimuli

When a single relevant stimulus (target) is presented among several irrelevant ones (odd stimuli), this stimulus must be selected among all the other ones. This selection process requires that attention is oriented toward the relevant stimulus while ignoring the odd ones.

In general terms, attention can be automatically captured (e.g., Posner and Cohen, 1984), or voluntarily oriented (e.g., Posner et al., 1980)¹. The distinction between automatic and controlled processes has been theorized and empirically evaluated by the pioneering work of Schneider and Shiffrin (1977) and Shiffrin and Schneider (1977), and has revealed to be particularly useful in several research fields and especially in the field of attention. Several properties are assumed to oppose these two modes of processing: while automatic processes are supposed to be “fast, parallel, almost effortless . . . not limited by capacity . . . not under direct subject control” (Schneider et al., 1984, p. 1), controlled processes present the opposite properties. Regan (1981) has pointed a very important distinction between two meanings of the term “automatic”: involuntary, [i.e., unintentionally triggered and, when triggered, impossible to stop intentionally (Kahneman and Treisman, 1984)] and effortless [i.e., capacity-free and not subject to interferences (Kahneman and Treisman, 1984)]. Regan noted that these two properties may not necessarily be tied (Regan, 1981).

Regarding attentional processes, the important point lies in the “costless” nature of automaticity since, according to Schneider and Shiffrin (1977), it is admitted that a “special type of automatic process” can “direct attention automatically to a target stimulus” (Schneider and Chein, 2003, p. 527).

An example of this automatic capture can be found in the “pop out” phenomenon. Because elementary features of a stimulus are processed in parallel, if a target differs from odd ones by a single elementary feature, then, attention will be automatically captured at its location. As a consequence, the RT or the error rate will remain unaffected by the presentation of a target among odd stimuli, whatever their number (Treisman and Gelade, 1980; Nakayama and Silverman, 1986)². In the visual modality, for example, these features can be color, orientation, brightness, size, depth, or direction of motion. Pop out effects can be accounted for by the fact that the visual structures process elementary features in (rather) separate specialized pathways (e.g., Livingstone and Hubel, 1988; Kastner et al., 1997; Treisman and Kanwisher, 1998, for a review).

Now, if the conjunction of two (or more) elementary features is needed to characterize a target among odd stimuli, the elementary features of the stimuli need to be bound to form unitary objects; attention would be responsible for this binding mechanism (Robertson, 2003), as metaphorically proposed by Treisman

and Gelade (1980) “. . . focal attention provides the “glue” which integrates the initially separable features into unitary objects” (p. 98). In this case, voluntary attention must be directed serially to each stimulus of a display which results in a linear increase of RTs with the increasing number of odd stimuli, (Treisman and Gelade, 1980; Nakayama and Silverman, 1986). In this case, odd stimuli act as distracters.

This distinction between automatic and controlled modes of attention orienting is not only justified by the psychophysical properties of human performance; it is also supported by physiological data showing that the way structures involved in attention orienting (namely prefrontal and parietal areas) are recruited, differs in the automatic and controlled modes (e.g., Buschman and Miller, 2007).

From what precedes it appears that whenever possible, it is preferable to present information in such a way that attention is automatically oriented toward the relevant stimuli. Moreover, when this is not completely possible, the number of stimuli to be processed at a time should not be too high, especially when one has to learn new skills. Although this last point may seem trivial it is not always born in mind in man-machine interface designs or man-machine interactions. Operators are often presented with several stimuli corresponding to information that *might* be useful, even when these stimuli are useless most of the time (the implicit idea here seems to be that any possibly useful information must *always* be available and presented to the operator); as a consequence, if not necessary at a given moment, these stimuli will act as distracters, especially for non experts during task learning. This, for example, might be the case in certain head up displays.

Let us now consider reading: there is no time pressure *stricto sensu* in reading but most readers do it at the fastest pace compatible with a good comprehension of the text. Learning to read is a long and difficult process; it seems that for these reasons, textbooks for learning to read have taken into account the aforementioned principles, maybe intuitively or by trials and errors, but in any case, most often, each page of these textbooks is very accurately organized. First, pages present, (1) very large letters (allowing easy and fast processing), (2) a few number of relevant information by page (which avoids too much attention sharing) and (3) they present the to-be-learned letters or association of letters in such a way that they pop out. Regarding this last point, for example, if the phoneme corresponding to a given letter (or to a group of letters such as “sh”) is the one to be learnt, then, the corresponding letter(s) is (are) generally written in bold or even in color, inside the word. As a consequence, the attention of the child is automatically (and effortlessly) driven to the relevant place(s) in the page and in the word.

These examples illustrate how simple principles regarding the organization of the stimuli to be processed may facilitate stimulus-related processing and, as a consequence, learning and performance.

RESPONSE-RELATED

Position of the effectors and response programming

Most often, when responses are triggered under time pressure, they are executed in a ballistic mode. When these responses are well learnt, it is often assumed that they are executed

¹Automatic capture and controlled orientation may also be referred to as “bottom up” and “top down” control of attention, respectively, in the literature (e.g., Buschman and Miller, 2007).

²Although more complex features (such as closure), or even extensive practice may result in “pop out” effects (reviews in Treisman, 1996; Hochstein and Ahissar, 2002), which stress, in the latter case, the importance of learning.

via motor programs (Paillard, 1960). These programs can be viewed as sets of abstract instructions specifying explicitly certain characteristics of the responses to be produced (e.g., Keele, 1968; Requin et al., 1991). Several behavioral (Rosenbaum, 1980; Lépine et al., 1989) and physiological data in monkeys (Georgopoulos et al., 1986; Riehle and Requin, 1989; Riehle, 2005) or humans (MacKay and Bonnet, 1990; Leuthold and Jentzsch, 2009) suggest that these programs specify certain parameters of the incoming movement.

Except in certain specific cases where they are innate, motor programs emerge from practice. In other words, most motor programs are learnt and, as such, the issue of motor programs is relevant to motor expertise and learning.

A consequence of the concept of motor program is that once the execution of the program has begun, it goes on until its end. A consequence of the parametric conception is that, for the parameters to be efficiently set before response onset, the initial state of the moving effectors must be known accurately. It has been established for long that, for programmed responses, proprioceptive/tactile signals are poorly involved in the control of ongoing activities but that they are critical to build, consolidate or update motor programs, during practice (Keele, 1973). Moreover, proprioceptive and/or tactile information is critical for being informed (before the movement) on the initial position of a given effector (Ghez et al., 1990; Gordon et al., 1995) and it has been shown that inaccuracy of initial proprioceptive signals results in pointing errors (Vindras et al., 1998). This indicates that this type of information is critical to set the appropriate parameters of the motor programs.

The quality of proprioceptive/tactile information is not uniform across all limb positions (Rossetti et al., 1994). Fortunately, it is possible to easily identify optimal positions since those which are spontaneously judged as the most comfortable are also those which produce more accurate position signals (Rossetti et al., 1994). These comfortable positions are those which correspond to the median range of each joint position. This median range also corresponds to a minimization of medial muscle tension (Rossetti et al., 1994). It is likely that "... discomfort could ... prevent the motor system from adopting postures where position signals are degraded, in order to keep the arm within a range of reliable encoding of posture" (Rossetti et al., 1994, p. 131).

As a consequence, a way to improve motor programs learning and/or motor programs execution is to improve the quality of initial proprioceptive and/or tactile information. Special attention must therefore be drawn on this point during the acquisition of motor skills. This is especially critical in young learners since proprioceptive information is less accurate in 6–8 children, as compared to 10–12 children: estimation of their static (initial) state is worse in the former than in the latter, which results in increased movement trajectory variability (King et al., 2012).

Although no clear explanation could be provided at that time, an example of application of these principles can be found in the famous method for keyboard playing by Bach (1753). Among the recommendations made by this musician, several concern the positioning of the arm, hand and fingers. Adopting these correct positions is critical for the future skills to be acquired as, according

to Bach (1753), incorrect positions will never allow the learner to play correctly. The teachers should make the learner adopt comfortable or natural positions especially (1) with the forearm slightly lower than the keyboard (which corresponds to a median position for the wrist articulation), (2) with avoiding large movements of the arm (which would involve several articulation which errors of position are additive: Rossetti et al., 1994), which should move as little as possible (3) with adopting positions for which the fingers are flexed and not stretched; these flexed positions (which, as can be understood from Bach (1753) correspond to median positions of the finger articulations) should allow the muscles not to be stiff, stiffness being an obstacle to accurate, smooth and adequate playing (remember that medial positions have also the virtue to minimize medial muscle tension: Rossetti et al., 1994). Moreover, still according to Bach (1753), a negative side effect of fingers stretching is that the position of the thumb remains too far from the other fingers (in other words, the articulation of the thumb is in an extreme position, which is inappropriate for accurate position information). One can imagine that more systematic attention should be paid to these aspects for other motor skills acquisition such as sport skills.

Moreover, in man-machine interfaces, several sensorimotor tasks have to be performed by operators (for example, push as soon as possible a remote button in response to visual or auditory information – which is a prototypic pointing task). No care is usually taken regarding the relative position of the buttons as compared to the usual resting position of the hand. No much attention is paid on the possible (or recommended) resting/average positions for the arms and hands. Therefore, the operators cannot choose the most appropriate positions which are imposed by the task and the interface.

Sequential movements, motor programming and chunking

Motor programs are learnt and, for sequential movements (such as playing music), once they are initially learnt, even more learning can improve them.

This is (at least in part) assumed to be achieved through clustering together sets of elements of the sequence (e.g., Sakai et al., 2003). This clustering operation is also called "chunking." Since activating motor programs takes time, chunking presents the advantage to pool together the elementary programs corresponding to each response of a sequence into a single program unit. Such an advantage is illustrated in the following example: in a simple RT task, although triggering a simple four-element finger movement sequence takes 62 ms longer than triggering a one-element simple finger response on a first day, this effect vanishes after 8 days training (Klapp, 1995). This effect of learning is explained in terms of chunking. When no choice is to be performed (simple RT task), subjects can choose and program the first element of a motor sequence before the onset of the response signal (RS; Klapp, 1974; Rosenbaum, 1980)³; as a consequence, the time necessary to program this first element does not contribute to RT. However, the other elements of the sequence must be programmed during the RT (and the time needed for this operation will increase the RT: Sternberg et al., 1978) and/or during the execution of the sequence,

³We will justify this assumption in the "intrinsic factors" section.

which may slower it. Therefore, chunking is a very efficient way to “compress” motor programs in order to save time when they are to be activated quickly. This cannot be done without training.

This interpretation has received support from physiological studies since, for example, the basal ganglia play a special role in chunking. Indeed, it has been shown that although it is possible to learn new sequences with basal ganglia impairment, chunking is no longer possible (Boyd et al., 2009; Tremblay et al., 2010).

Once again, it looks like keyboard players understood these principles, long before neuroscientists, since different learning methods seem to take chunking effects into account. Playing a keyboard is typically a sequential activity and, for example, Hanon (1873), in his famous “Le pianiste virtuose . . . (The virtuoso pianist . . .)” compilation of 60 exercises, recommends, when beginning to learn, to play one’s scale by stressing one (or two) note(s) every, say, 5–6 notes. This, of course produces a rhythm which is not actually in the score but, he says, this will facilitate learning. This facilitation can be interpreted in terms of chunking facilitation. Indeed, it is clear that these rhythms comprising 5–6 notes create artificial structures in the scale and allow grouping elements into chunks, which, as we mentioned earlier, is very efficient in term of fast sequence programming. We can see here that Hanon (1873) recommends (counter-intuitively) not to begin learning his exercises as they should be played; on the contrary, he takes into account first, what is better for learning and, only in a second step, what is desirable for playing.

Moreover, as indicated earlier, the activation of motor programs takes time. This is certainly why, Hanon or Bach recommend playing their exercises much slower than required on the score and, as learning goes by, to accelerate accordingly. This is a very efficient way to take into account that, with training (because of chunking), the number of to-be-programmed elements will decrease; as a consequence, the time needed to program (more exactly to activate the motor programs of) these elements will also decrease and will no longer be an obstacle to fast playing. Bach (1753) writes that by accelerating progressively, the finger placement will become so fluid that there is “*no need to think of it.*” In our terms one could translate (less artistically) the preceding words by: the finger movements of the different sequences of the piece are “*fully programmed.*”

RESPONSE SELECTION

Even when stimuli can be identified effortlessly and when responses are very easy to execute, certain sensorimotor tasks may be very difficult to perform because certain stimulus-response associations (or, in other words, response selections), are very difficult to implement.

Response selection refers to determining which response alternative is required by the RS, and must be distinguished from response programming (more exactly motor program activation), which, as we just discussed in the preceding section, refers to organizing the selected movement. This determination process is, therefore, neither directly dependent on the stimuli, nor directly dependent on the responses and unit recordings in monkeys indeed evidenced neurons reacting neither to a response nor to a stimulus but selectively reacting to specific stimulus-response

associations (Zhang et al., 1997). This process must rely on a stimulus-to-responses (S-R) mapping. This mapping will define the nature of the task and, of course, it is most often established by instruction. This last point is noteworthy since it is relevant to the issue of learning: a learner who is taught a task by his/her teacher will (in principle) comply with the instruction he/she is given.

Rule-based vs. list-based S-R mapping

There are (at least) two ways to establish a mapping relationship between a stimulus set and a response set: (1) a rule-based S-R mapping and (2) a list-based S-R mapping.

In case (1), a rule establishes the nature of the S-R mapping; for example instruction requires producing a right response to the presentation of even digits and a left response to odd digits. In case (2), instruction consists in an arbitrary list of digits-to-finger mapping; for example 0, 3, 4, 6, or 7 require a right response and 1, 2, 5, 8, or 9 a left response. In case (1), the difficulty of the response selection process will depend on how easily this rule will be applied. In case (2) the difficulty of the response selection process will depend on the length of the list (Hasbroucq et al., 1990; Hasbroucq, unpublished). All other things being equal, it is easier to apply and learn a rule-based mapping than a list-based one (Hasbroucq et al., 1990; Hasbroucq, unpublished).

Response uncertainty has a very strong impact on performance. This can be experimentally manipulated by varying the number of response choices; it is well established that the RT and the error rate increase with the number of possible responses (Hick, 1952). Of course, the number of choices can affect each level of information processing: the perceptual level if, for example, uncertainty involves the different positions of the possible RSs, as well as the response level since only one among several possible response programs must be activated.

Regarding the response selection level, the effect of the number of alternatives depends on the nature of the S-R mapping: rule-based or list-based. It is clear that when list-based, the difficulty of the selection process will mainly depend on the size of the list from which the subject must retrieve, at each trial, the response corresponding to the presented stimulus. On the contrary, the process of applying a rule, being only dependent on the nature of the rule, rule application is not supposed to depend on the number of choices (Hasbroucq et al., 1990) and empirical evidence support this view (Hasbroucq, unpublished). Therefore, teachers, task prescribers or man-machine interfaces designers should be aware of these principles and try to apply them whenever possible.

Stimulus-response compatibility effects

Stimulus-response compatibility (SRC) effects can be defined as “... the modification of performance induced by a change in the SR [mapping] relationship that is uncorrelated with any change in stimulation or responding” (Hasbroucq and Guiard, 1991, p. 246). This definition is not theoretic but empiric and does not refer to any specific mechanism. However, since this type of effects cannot be accounted for by purely stimulus-related or purely response-related phenomena, it must be assumed that it affects response selection processes (e.g., Sanders, 1990). From this definition,

it also appears that SRC effects only have to do with rule-based mapping. Let us consider a very simple case of between-hand choice RT task. Suppose that the stimuli consist in a visual dot presented on the right or on the left side of the subject. If subjects are instructed to respond on the same side as the stimulus (right and left responses to right and left stimuli respectively), the RT is shorter (Shaffer, 1966) than if subjects are instructed to respond on the side opposite to the stimulus (right and left responses to left and right stimuli, respectively). This simple case of spatial SRC effect indicates that, all other things being equal, certain associations between stimuli and responses are easier to apply. These concern instruction-based relationships.

Stimulus-response congruency effects

Now, it seems that irrelevant S-R transformations can also proceed automatically and influence RT, because overlearned. An example of these automatic, irrelevant, overlearned S-R transformations may be found in the Simon task (Simon, 1990; Lu and Proctor, 1995, for reviews). In a Simon task, subjects have to respond spatially (e.g., right or left; up or down ...) to a non spatial feature of a RS (e.g., color, pitch ...), while ignoring its spatial irrelevant feature. For example, subjects have to give a right response to a blue stimulus or a left response to a yellow one, regardless of the stimulus position (right or left). When the position of the required response and that of the stimulus correspond (congruent condition) the RT and the error rate are smaller than when response and stimulus positions are opposite (incongruent condition).

Although it is not warranted that the Simon effect involves response selection processes only (Hasbroucq and Guiard, 1991), response selection may be influenced by congruity (Carbonnell et al., 2013).

It seems that, in a Simon task, two S-R transformations are activated in parallel: a controlled instruction-based transformation plus an automatic one (always congruent, established by previous experience or, in other word, learning)⁴.

On congruent conditions, both the automatic and controlled S-R transformations activate the same (correct) response. On the contrary, on incongruent conditions, while the controlled S-R transformation activates the correct response, the automatic transformation activates the incorrect one. As a consequence, on incongruent conditions, these two S-R transformations will compete and the resolution of this competition (Kornblum et al., 1990) will take time (which increases the RT) and may sometimes fail (which results in increased error rate). In more complex situations, *a priori* naive judgements are not sufficient to determine which associations will be congruent and which ones will not (Payne, 1995; Vu and Proctor, 2003; Hoffmann, 2010).

Now, what learning has done (establishing automatic associations), learning can also undo it. If subjects are trained to perform a spatially incompatible RT task (i.e., respond right or left to left

or right stimuli, respectively), and have to perform a Simon task after, the Simon effect vanishes (Tagliabue et al., 2000, 2002).

Before practice, tasks are often explained to learners by instruction; instruction being usually the first event when teaching a task. Instructions alone can establish or abolish stimulus-response association without practice. If subjects must pronounce the meaningless syllables “bee” or “boo” in response to the presentation of words meaning either right or left and, in other trials, must give a “bee” or a “boo” response to blue or green squares, respectively, RT is faster when the blue square is presented to the right than when it is presented to the left: an instruction-based Simon effects appears (De Houwer et al., 2005). Conversely, if a preparatory instruction indicates which mapping (compatible or incompatible) should be applied for a spatial task following a Simon task, in the same trial, the regular Simon effect vanishes on the (first) Simon task, if the instructed mapping of the spatial task is incompatible (but persists if the spatial task mapping is compatible; Theeuwes et al., 2014).

To sum up: (1) certain S-R mapping rules, equally easy to understand (e.g., respond on the same side as the stimulus, or respond on the side opposite to the stimulus), are not equally easy to implement; (2) given a S-R mapping rule (e.g., respond left to blue stimuli or right to yellow ones), certain S-R associations may facilitate (congruent associations) or impair (incongruent associations) response selection processes and, as a consequence, performance; (3) the congruent, incongruent (or neutral) nature of a new S-R association cannot be determined naively but must be established empirically, (4) the way learners are instructed to perform a task has a critical influence on response selection processes (e.g., Tandonnet et al., 2014) since, (a) instructions establish S-R associations, (b) instructions alone can induce congruency effects, (c) instructions alone can abolish congruency effects.

INTRINSIC FACTORS

Dependent on the structure of tasks, advance information regarding the to-be-performed responses is available or not. This prior information may allow subjects to orient appropriately their attention, to select in advance the appropriate response, to program completely or partially their response. Of course, this depends on the structure of the task and, as such, availability of advance information belongs to extrinsic factors. Now, subjects can decide to use or not to use advance information and, as such, the effects of advance information also depend on intrinsic factors. Because, the effects of advance information as well as “purely” intrinsic factors can be unified in terms of executive control, we chose to discuss them in this section.

PREPARATION

Advance information is supposed to reduce subjects’ uncertainty regarding forthcoming events. This information can be relative to events proper (for example, the nature of a RS). This information can also be relative to timing (for example the moment of occurrence of a RS). Therefore, when prior information is available, it is possible to get prepared, either to *which* event(s) will occur or to *when* event(s) will occur, or both. This preparation can be evidenced by the fact that RT is much faster when preparation

⁴Remembering the distinction between “involuntary” and “effortless” meanings of the term “automatic” (Regan, 1981), the important point here is that the congruent response is triggered involuntarily since “once learned, an automatic process is difficult to suppress, to modify, or to ignore” (Schneider and Shiffrin, 1977, p. 2).

is possible than when it is not. Depending on the nature of advance information (regarding “what” or regarding “when”), Requin et al. (1991) distinguished two, not mutually exclusive, types of preparation: event preparation and time preparation.

Event preparation

If a soccer goal-keeper facing a penalty shoot moves toward one side before the shooter kicks the ball, this information will be used by the shooter to score on the other side. Now, if the goal-keeper waits for the kick and, then, tries to react appropriately, he/she will never stop the ball because his/her RT will be too long. Therefore, goal-keepers make a bet, “choosing” one side in advance and, if the shooter happens to shoot at this same side, they get a chance to stop the ball. What do journalists actually mean when saying that a goal-keeper who stopped a penalty shoot has “chosen” the right side? They mean that (according to which his coach taught him), he/she selected one side and prepared a jump toward this side. As a consequence, his/her RT has been short enough to allow him/her having his/her hand at the right place on time, and stop the ball⁵. This example illustrates the benefits of event preparation in the domain of response preparation.

Responses. But what do we mean when we say that the goal-keeper has “prepared” a jump in a predetermined direction? An important observation is that event preparation reduces RT. Therefore, either processing speed regarding some operations increases when preparing or, at constant processing speed, certain operations are done in advance and are not required anymore after the RS, which reduces RT.

First, the goal keeper can, at least, decide in advance toward which side he/she will react. In other words, the selection process can be achieved before the ball is kicked and this contributes reducing RT. But what about motor programs?

When a response is programmed, it has necessarily been selected. Therefore, selection and programming operations may often be confounded factors. To disentangle selection and motor programming, a solution proposed by Klapp et al. (1974) is to evaluate motor programming while maintaining constant the number of choices. Klapp et al. (1974) wondered whether the duration of a motor response could be a specified parameter of motor programs. In a RT paradigm, subjects had to produce either a short (150 ms) or a long (600 ms) keypress. In choice RT conditions (where no program can be prepared), RT was shorter before a short than a long keypress. On the contrary, if a cue indicated before the RS which response (short or long) should be produced, no RT difference was found between short and long responses. The authors therefore concluded that the short–long differences in choice RT revealed differences in the times needed to program a short and a long response. The absence of such an effect when the same responses were precued indicated that, in this case, programming operations had taken place before the RS, and, consequently, did not influence RT anymore. This interaction between response duration and conditions (uncued *vs* precued response duration) was a strong

argument in favor of the idea that, with prior information, motor programming can occur in advance. In other words, what has been done during the preparatory period (PP; programming) has no longer to be done after the RS (which contributes reducing RT). Note that, in Klapp et al.’s (1974) paradigm, there was no confounding factor (e.g., selection) with response programming. The short–long effect has been reproduced several times by different teams (e.g., Zelaznik and Hahn, 1985; Vidal et al., 1995).

Klapp et al. (1974) inferred from the RT pattern they observed that response duration was prepared before the RS when precued. Vidal et al. (1995) provided physiological support in favor of this view. They recorded electroencephalographic (EEG) activity over motor areas in humans during the PP of a precueing (Rosenbaum, 1980) paradigm where subjects had to produce either a short (700 ms) or a long (2500 ms) interval delimited by two brief button presses. Two seconds before the RS, a precue could either indicate which response (short or long) should be produced, or give no information regarding response duration. The short–long effect was reproduced. Moreover, during the PP, EEG activity was small when the cue gave no advance information, and larger when the cue indicated which response to produce. However, this simple effect could be attributed to motor programming, response selection or both. The important finding was that, when the cue indicated which response to produce, EEG activity was larger for the short than for the long response during the PP. In other words, when duration programming was assumed to occur during the PP, a short–long effect was observed on the amplitude of preparatory EEG activity. It is to be noted that this effect was observable over the supplementary motor areas (but not over the primary motor areas) in the early part of the PP, while the same effect showed up in the very late part of the PP over the primary motor areas (but not over the supplementary motor areas anymore). This latter point suggested that duration programming had taken place (at least in part) in the supplementary motor areas with advance information and that the program had been later transferred to the primary motor areas, just before the “go” signal, for execution.

Unit activities recordings from monkeys also support the view that response parameters can be programmed in advance. For example, Riehle (1991) could record primary motor cortex neurons which activity was phasically related to the precue when this signal indicated in advance which response was to be executed and these neurons did not react after the RS. On the contrary, when the precue gave no advance information regarding the response, these neurons were almost inactive after the precue but phasically discharged after the RS (which, in this case, gave all information regarding the response to be produced). Once again, the activity of these neurons suggests that what had been done before (shortly after the precue), has no longer to be done after the RS; with uninformative precues, this still needed to be done just after the RS.

Coming back to the goal keeper, it seems quite safe to conclude that, when \ll choosing \gg a side, he/she selects this side first and programs all the initial sequence of his/her jump. These two processes do not contribute to his RT anymore; as a consequence, his RT is shorter, which increases his/her chances to stop the ball.

⁵In this case, it is likely that the side of the jump is chosen in advance while the hand (or foot) stopping movement is determined after.

Stimuli. Preparatory information regarding events may also improve operations upstream programming and selection processes, that is, stimulus processing. This has been clearly and elegantly demonstrated by Posner et al. (1980). In a RT task, subjects had to respond as soon as possible by a keypress after a RS which could be presented to the right or to the left of a fixation point. Before the RS, an arrow pointing to the right or the left indicated on which side the RS was more likely to occur: in 80% of the trials, the RS occurred on the side indicated by the arrow (valid trials) and in 20% of the trials, the RS occurred on the other side (invalid trials). Although subjects had to keep their gaze oriented on the fixation point, RT was shorter in valid than in invalid conditions. The important point to be noticed, here, is that the response to be produced was always the same. Therefore, the decrease in RT could not be attributed to advance selection or programming processes. Posner et al. (1980), interpreted their results in term of attentional effects: covert attention was oriented on the side indicated by the arrows. This allowed faster detection of the stimuli in the valid condition, at the cost of slower detection (as compared to a neutral condition) in the invalid condition. These effects have been reproduced several times ever since (e.g., Coull and Nobre, 1998). They demonstrate how voluntary orientation of attention improves stimulus processing, provided that attention is appropriately oriented.

Time preparation

At Atlanta's 1996 Olympic Games, British sprinter Linford Christie false-started twice in the men's 100 m final and was disqualified from the competition. He refused the decision, remained on the track and delayed the final for several minutes. At Paris's 2003 world championship, American sprinter Jon Drummond also false-started in a 100 m quarterfinal. Drummond also contested the decision and stayed on the track repeatedly shouting "I did not move."

What happened to these champions illustrates an interesting aspect of time preparation. Before a 100 m race, there is no uncertainty regarding the "what," which can be fully prepared. Preparing the "when" would also reduce RT but, since the delay between the "get set" order and the "go" signal is *a priori* unpredictable, it seems that time preparation is impossible in this situation. However, since this delay cannot last very long, as time goes by after the "get set" order, the probability that the "go" signal will occur "right now" increases progressively and the level of preparation may increase accordingly. One can therefore imagine that these two sprinters, under strong motivation, prepared as well as possible the motor sequence corresponding to their start but also got prepared for the moment when the "go" signal would occur, so much prepared that refraining moving began impossible too early; they probably did not feel that they had anticipated the go signal (which, it seems, they actually did).

The case of the abovementioned sprinters is particular because they prepared their move and (to a certain extent) the moment to move. Although counter intuitive, it is also possible to prepare the moment without being able to prepare the response. For example, in choice RT tasks in which the moment of occurrence of the RS is predictable, RT is faster than when the occurrence of the RS is not

(or less) predictable (Requin et al., 1991, for a review). However, the locus of the effect of time preparation is still a matter of debate. In the following, we will provide arguments indicating that time preparation affects (at least) sensory and motor processes⁶.

Responses. Absolute accuracy of time estimation decreases as the duration to be estimated increases (Gibbon, 1977). As a consequence, preparation is better timed for short foreperiods than for longer ones. This would explain why RT increases with increasing foreperiod duration⁷, provided that each foreperiod duration is administered in different blocks of trials (Woodrow, 1914). Therefore, a convenient way to manipulate time preparation is to manipulate the duration of PPs across blocks of trials.

It has been shown that time preparation modifies the excitability of corticospinal pathways during the PP (e.g., Davranche et al., 2007). However, these preparatory effects do not demonstrate that motor processes are speeded up. Therefore, Tandonnet et al. (2003, 2012) examined the time course of activation of the primary motor cortex during the RT period following short or long foreperiods, with EEG and transcranial magnetic stimulation (TMS). In a between-hand choice RT task, Tandonnet et al. (2003), used Laplacian-transformed data which allowed them to examine EEG activities of the primary motor cortex contralateral to the responding hand (M1). The authors showed that the time separating M1 activation from EMG onset was shorter after a short (500 ms) than a long (2500 ms) foreperiod. This indicated that better time preparation (during short foreperiods) speed up motor processes. In other words the authors showed that there is (at least) a motor locus for the effects of time preparation. In the same vein, Tandonnet et al. (2012), using TMS, examined the time course of excitability of the primary motor cortex contralateral to the responding hand in a between-hand choice RT task following the same foreperiods. The authors showed that, after the RS, the amplitude of the motor evoked potential of the responding hand increased earlier following the short than the long foreperiod. Note that such an effect was absent for the non-responding hand. Moreover, the dissociation between responding and non responding hands occurred earlier after the short than after the long foreperiod. This again indicated that the required response is implemented faster in the corticospinal system when time preparation is more efficient. As a consequence, it seems that time preparation speeds up motor processes (even when event preparation is impossible), which contributes improving performance.

Stimuli. Time preparation can also improve stimulus processing. Coull and Nobre (1998) examined the effect of time preparation on stimulus processing using a temporal variant of the Posner et al.'s (1980) task. In a simple RT task, subjects had to respond as soon as possible by a keypress after a RS which could be presented to the right or to the left of a fixation point. Before the RS, a preparatory signal could prime the duration (short: 300 ms or

⁶Although effects of time preparation on central processes cannot be excluded, this idea is, to our opinion, less grounded yet; therefore we will not present or discuss it here.

⁷This effect holds for foreperiods superior to 200 ms otherwise, there is no time enough to get prepared (Bertelson, 1967).

long: 1500 ms) of the foreperiod (i.e., the moment when a RS would be presented). This prime was valid in 80% and invalid in 20% of the trials. RTs were faster for validly than invalidly primed foreperiods. However, this effect was much larger for invalid long primes and modest (or even absent) for invalid short primes. The authors convincingly argued that when subjects have anticipated a long foreperiod, the RS occurs too early and finds subjects in an unprepared state, which impairs their performance. On the contrary, when subjects have anticipated a short period and the stimulus has not occurred after a time corresponding to the end of the short foreperiod, subjects necessarily know that the stimulus will occur later, at the end of the long foreperiod. In this case, they can reorient their attention during the waiting delay, which allows them maintaining their performance.

Note that, in this experiment again, only one response was possible; therefore RT variations could not be attributed to selection and/or programming operations. Subject only had to detect the signal and, then, execute the pre-selected and pre-programmed response. Now, keeping in mind that time preparation can speed up motor processes (Tandonnet et al., 2003, 2012), it could well be that the RT advantage provided by time preparation was in part or completely due to motor improvement (speeding up). However, Davranche et al. (2011), in a detection task without any time pressure, demonstrated that time preparation actually facilitates stimulus detection for precued short foreperiods.

Coming back, now, to false start 100 m races, it is clear that, although accurate event preparation is encouraged, it seems that time preparation is prohibited. Indeed, Linford Christie was disqualified not because he moved before the go signal but because he reacted too fast (60 ms). RTs inferior to 100 ms are considered as false starts because it is assumed that reacting in less than 100 ms is not physiologically possible. Let us indicate here that we feel that this opinion is not firmly physiologically grounded and appears somehow arbitrary. If one really wants to discourage time anticipation, there are psycho-physiologically efficient ways to avoid it. Instead of having the go signal given by a human operator, it could easily be given by an automatic mechanism using so called “non aging” probability functions to determine the occurrence of go signals. Such a procedure would render almost impossible time preparation and, therefore, would probably avoid these recurrent false start problems.

To sum up, as soon as any type of information is available, whether it concerns the nature of the incoming events or the moment when something will occur, the human brain adapts in advance. By doing so, it anticipates future events thereby avoiding that adaptation takes place under time pressure, in reaction to these events. In other words, preparation processes can help release time pressure since all computations done in advance don't need to be repeated during the time left to react. Therefore, when relevant events occur, the “reactive” processing cost is lower thereby increasing processing efficacy. Several examples of anticipatory adaptation to predictable events can be found in other domains of physiology such as blood pressure regulation, thermoregulation or regulation of glycemia.

Event and time preparation are of prominent importance for several activities that must be learnt such as, for example, driving, sport, hunting, or music.

In driving, a large part of teaching consists in making learn what must be anticipated. On the event side of preparation one can cite (non exhaustively): learning to get prepared to the nature of the road given the corresponding road signs, get prepared to the reactions of the car to the drivers' actions, get prepared to the possible moves of the other drivers . . . On the time side of preparation one can cite (non exhaustively): learning to get prepared to the time it takes to stop after breaking, get prepared to the moment when the traffic light will turn red/green, get prepared to the deceleration in a very close future of the preceding vehicle when the braking lights turn on . . .

In a similar way a hunter, before becoming a good hunter, must learn to read (interpret) the signs available in nature which allow him/her to anticipate and get prepared to what the game might do and when it might to it.

In team sports, a large part of learning also consists in becoming able to “read” the game. Reading the game amounts to being able to anticipate the moves of the other players, get prepared and act quickly in consequence. This preparation relates, not only on *what* the other players will do (or will not do) but also relates on *when* they might do it. In certain sports, such as rugby football, certain game sequences are completely repeated and learnt before the matches. This training will not make the players, say, run faster. However, it will allow accurate execution of motor programs (see preceding section) and, overall, full preparation of each player to the different events of the sequence. This full preparation gives them a significant advantage as compared to the adverse players who cannot do better than react to this sequence – except if the adverse coach has studied videos of previous matches and trained his own team to anticipate for such sequences (if they are used too often). Anticipation is also critical in other sports such as combat sports or any sport in which, reading and anticipating opponent's actions provides an serious advantage (e.g., tennis).

Finally, in music, temporal preparation is critical: anticipation of rhythmic patterns is essential for musical expectancy and, of course, for accurate performance. Moreover playing instruments presenting a certain “inertia” (for example there is a non negligible delay between the moments when a musician blows in a tuba and the moment when the corresponding sound is produced) needs that the musician learns (and is prepared to) this inertia before being able to play in an orchestra. Event preparation is also important and must be learnt in music. When playing together, musicians performing music with a sketchy outline voluntarily exchange non-verbal cues; these cues act as preparatory signals and can be non temporal (specific movements, face expressions, glances . . .) or temporal information. For example, jazzmen, in their communication, learn to exchange temporal cues which take the form of subtle deviations from the well-established rhythmic pattern (Vuust et al., 2005).

STRATEGIC EFFECTS: SPEED-ACCURACY TRADE-OFFS

Among strategic effects SATs are well documented. As we will see below, it is possible to trade speed against accuracy and accuracy against speed. Long term (between experimental conditions) strategic changes are called macro trade-offs, while short term (from trial to trial) changes are called micro trade-offs (e.g., Jentzsch and Leuthold, 2006).

Macro trade-offs

As indicated in the first section, increasing time pressure by constraining the time let for giving the appropriate response after a RS, results in an increase in error rate. This is an extrinsic constraint. Now, subjects can also choose to increase speed or accuracy spontaneously or, for example, to comply with experimenter or teacher instructions. If they increase speed, this will be at the cost of increasing the error rate. Conversely if they choose to increase accuracy, this will be done at the cost of increasing RT. In other words, subjects can trade accuracy against speed and conversely. This trade-off takes the form of an S-shaped curve. This phenomenon has been called SAT (Fitts, 1966; Pew, 1969). This phenomenon can be used for training and, according to the performance pattern displayed by learners, teachers can encourage their trainees to favor speed or accuracy at different stages of learning and performance. Indeed, subjects comply quite easily with speed/accuracy instructions.

Micro trade-offs

In choice RT time tasks, as mentioned above, errors may occur. RT of errors is often (but not always) faster than that of correct responses (Smith and Brewer, 1995). After errors, the RT is longer (post-error slowing) than after correct responses (Rabbitt, 1966; Smith and Brewer, 1995; Allain et al., 2009) and the error rate may also be lower (Laming, 1979; Ruitenberg et al., 2014). On the contrary, before errors, RT is shorter (pre-error speeding) than before a correct response (Smith and Brewer, 1995; Allain et al., 2009; Dutilh et al., 2012a). This saw tooth pattern of RT suggests that these sequential adjustments are strategic and reflect trial-by-trial micro trade-offs (Smith and Brewer, 1995). After an error, subjects would adopt a more cautious strategy (Laming, 1979; Dutilh et al., 2012b; Ruitenberg et al., 2014; but see Notebaert et al., 2009 for alternative accounts) and on the subsequent trials, subjects becoming more and more confident would progressively speed up until an error is generated again. These micro trade-offs suggest that an action monitoring system is at work at each trial and, as a function of performance, regulates responding behavior in order to optimize it (Botvinick et al., 2001). Therefore, these micro trade-offs would reveal trial by trial, “off line,” action monitoring.

The implication of post-error slowing in learning is not obvious; however, it is possible that this type of adjustment reflects a fast short-termed learning process from the preceding error.

ON LINE ACTION MONITORING

In the preceding section, micro trade-offs suggest that an action monitoring system triggers behavioral adjustments to optimize performance. These adjustments occurring in reaction to errors, we can qualify them as “reactive.” Of course, these reactive adjustments occur *between* trials. One could wonder whether such reactions to errors might also occur *within* trial.

Reactive action monitoring

EMG indices. In a between-hand choice RT task, Allain et al. (2004) recorded surface electromyographic (EMG) activity of the prime mover muscles involved in the two possible responses. They showed that the amplitude of the erroneous EMG bursts was smaller than that of the correct ones. The initial slope of these

EMG bursts being identical on correct responses and on errors, it is likely that the initial motor command was identical in both types of responses. Therefore, it seems that the smaller EMG bursts observed on errors reflect an inhibition of the motor command during its execution. If this interpretation is valid we can conclude that once the error is triggered, the action monitoring system detects it and reacts in an attempt to stop it. Now, considering that these errors were committed, it seems that the action monitoring system failed. What could, therefore, be the functional significance of this EMG effect?

Coles et al. (1985) showed that, in between-hand choice RTs tasks, certain correct responses are preceded by a small sub-threshold EMG activation (insufficient to trigger an overt response) on the “wrong” side. The RT of these correct trials is longer than that of correct trials which do not contain sub-threshold incorrect EMG activations (termed “pure” correct trials, in the following). These observations have been reproduced several times ever since (e.g., Allain et al., 2009).

There is a general agreement that, since these sub-threshold activations occurred on the “wrong” side, they correspond to partial errors. Burle et al. (2002b) proposed, in addition, that these partial errors correspond to almost errors that have been detected, inhibited and corrected on time. Allain et al. (2009) provided arguments in favor of this view, showing that sequential speeding and slowing adjustments also exist before and after partial errors respectively, although the size of these adjustments is smaller for partial than for full blown errors. Moreover, the fact that RT is longer on partial error trials than on pure correct trials could be a sign that, after a partial error, a part of processing operations must be done again in order to produce the correct response.

If these interpretations are correct, the existence of partial errors indicates that the action monitoring system is most often able to detect, inhibit and correct erroneous motor commands. When it works, this results in a partial error; when it fails this results in a full blown error; however, on errors, a sign of the tentative inhibition implemented by the action monitoring system can be found in the smaller size of the EMG bursts as compared to pure correct responses. Finally, still if these interpretations are correct, it is possible to calculate an index of the efficiency of the action monitoring system: the correction ratio, which corresponds to the ratio between successfully corrected incorrect activations (partial errors) and the total amount of incorrect activations (partial errors plus full errors; Burle et al., 2002b). For example, in the Simon task, the error rate is higher on incongruent than on congruent trials, as indicated above. However, a part of this effect is due to a decrease of the correction ratio on incongruent trials. Therefore, a part of the increase of the error rate on incongruent trials is due to a lowered efficiency of the action monitoring system. This indicates that there are two (not mutually exclusive) ways to deteriorate performance in terms of accuracy: decreasing the efficiency of the information processing chain, or decreasing the efficiency of the action monitoring system (or both, as it is the case on incongruent trials of a Simon task).

EEG indices. Falkenstein et al. (1991) discovered that a large response-locked event-related potential (ERP) was evoked by

errors. Since this ERP seemed specific to errors, it has been called error negativity (Ne) or, later on, error-related negativity (ERN: Gehring et al., 1993)⁸. Scheffers et al. (1996) showed that an Ne was also evoked by partial errors, although its amplitude was smaller than on errors. This observation makes sense if one admits that partial errors are almost errors. Finally, using Laplacian-transformed data, Vidal et al. (2000) unmasked a small Ne-like on correct trials, too, that had been unnoticed so far. It is clear that the discovery of the Ne constituted strong evidence for the existence of an action monitoring system able to quickly⁹ separate errors from correct responses at the very moment of the response. However, revealing the existence of an Ne-like on correct responses, also raised the question of the relationship between the Ne and the Ne-like waves: if the Ne-like is just a small Ne, then current models of action monitoring need to be revised to incorporate this finding that they cannot account for; if the Ne-like corresponds to another component, the Ne is specific to errors (full blown or partial) and there is no need for such a revision. Recent data (Bonini et al., 2014) suggest that the Ne and the Ne-like share (at least) a common source: the supplementary motor areas proper (SMAp). This finding suggests that the Ne-like and the Ne correspond to a same component which is modulated in amplitude: small on correct responses, larger on partial errors and even larger on full blown errors. Although this sensitivity of SMAp to performance is a sign of the existence of an action monitoring system, as initially shown by Falkenstein et al. (1991), its functional significance is not obvious. It has been proposed that SMAp could generate warning “default” signals which amplitude would depend on the ongoing performance (Bonini et al., 2014).

The sensitivity of SMAp to performance, just after EMG onsets, corresponds to another piece of evidence that the action monitoring system reacts *within* trial to the quality of performance.

Proactive action monitoring

In the preceding sections, we have seen that EMG and EEG data indicate that the action monitoring system *reacts* to incorrect activations in an attempt to correct them and avoid full blown errors. One could, finally, wonder whether the action monitoring system could *act before* incorrect activations occur in an attempt to prevent them.

Hasbroucq et al. (2000), using H reflex showed that, in a between-hand choice RT task, the excitability of spinal motoneurons corresponding to the responding hand increased, while it decreased on the other side, just before response execution. Burle et al. (2002a), using TMS showed that, in a between-hand choice RT task, the excitability of the primary motor cortex contralateral to the responding hand (contra M1) increased before response execution, while that of the primary motor cortex ipsilateral to the responding hand (ipsi M1) decreased. Finally, using Laplacian-transformed EEG recordings over contra and ipsi M1, Vidal et al. (2003) reported, in a between-hand choice RT task, a response-locked negative/positive pattern, likely corresponding to an EEG counterpart of the activation/inhibition pattern previously

evidenced with other techniques by Hasbroucq et al. (2000) and Burle et al. (2002a). While it had previously been shown in monkeys that the negativity observed over contra M1 corresponds to the activation of the apical dendrites of layer V of contra M1 (Arezzo and Vaughan, 1980), the nature of ipsilateral inhibition appeared less clearly. Considering that, before spontaneous movements, contra M1 activation is present but not ipsi M1 inhibition (e.g., Ikeda et al., 1995), Vidal et al. (2003) proposed that this inhibition was needed for preventing the emission of the “wrong” response. In other words, ipsilateral inhibition could represent the existence of an *a priori* mechanism of error prevention set proactively by the action monitoring system. In a go no-go task, errors are possible but, contrary to choice RT ones, not with the non responding hand. Vidal et al. (2011) showed that ipsilateral inhibition was absent in a go no-go task, which was in line with the interpretation of ipsi M1 inhibition by Vidal et al. (2003). Finally, in a between-hand choice RT task, Meckler et al. (2010) manipulated the risk of committing errors. They compared a “classical” choice RT with a biased one. In the classical condition, each response was equiprobable (50%) while in the biased one, one of the two responses was 80% probable (and the other one 20%, only). In the 80% condition, the response was expected, in the 50% condition there was no specific expectation, while in the 20% condition the response was unexpected. RT and error rates increased from expected to no expectation conditions and from no expectation to unexpected conditions.

Contra M1 activation was not affected by expectation. On the contrary, ipsilateral inhibition was small in the expected condition, larger in the no expectation condition and even larger in the unexpected condition. Meckler et al. (2010) concluded that ipsilateral inhibition was actually set to prevent errors, especially when considering that, in the unexpected condition, there was an inverse (between-subjects) correlation between the strength of ipsilateral inhibition and the error rate. In other words, in the condition where the risk of committing an error was highest, those subjects whose inhibition was stronger were those who committed the smallest proportion of errors.

Therefore, it seems that ipsi M1 inhibition is a physiological marker of a “double-checks” proactive action monitoring mechanism. Whether or not this proactive mechanism develops during the task by training is not known, yet. However, the possibility to develop this inhibition needs a certain degree of maturation. Van de Laar et al. (2012) showed that ipsi M1 inhibition was neither present in 8-years old nor in 12-years old children. It is worth noticing that the absence of inhibition in children was associated to almost 3 times (2.9) more partial errors and 3 times more errors than in 20-years old young adults. Although Van de Laar et al. (2012) did not address or discuss the following point, it is easy from their Table 2 (which reports partial and full error rates) to calculate correction ratios (Burle et al., 2002b). A striking result appears: young children corrected their incorrect activations in 75% of the cases while young adults corrected them in 76% of the cases. Although no statistical data are available in Van de Laar et al. (2012), it is very likely that correction rates of children and adults did not differ. As a consequence, it is not action monitoring as a whole which is deficient in children: their high error rate cannot be

⁸The Ne must not be confounded with the feedback-related negativity (FRN: Miltner et al., 1997) which may be elicited by error feed-backs.

⁹The Ne begins about 30 ms after EMG onset but before the mechanical response.

attributed to reactive action monitoring failure, as revealed by partial errors. It seems, on the contrary, that their deficiency must be found, at least in part, in immature proactive action monitoring, as revealed by their lack of ipsilateral M1 inhibition.

To sum up, recent research showed that, even under time pressure, sensorimotor information processing may not be completely unidirectional: reprocessing can quickly be triggered by the action monitoring system in reaction to incorrectly activated responses. These reactive mechanisms allow avoiding several overt errors. Moreover, not only does the brain react to incorrect activations but it also anticipates them and sets preventive mechanisms to avoid them in error-prone situations. It is noteworthy that the reactive side of the action monitoring system is efficient quite early in childhood, but that its proactive side needs a longer maturation to optimize performance in sensorimotor activities.

From what precedes it is quite clear that the human brain is not structured to function errorless, this having been recognized long ago by Seneca through his famous “*errare humanum est*.” What is newer is the fact that errors are largely dealt with at different levels of information processing: prevention, detection, inhibition, correction and, if these mechanisms finally failed, strategic behavioral adjustments after errors.

The intrinsic proneness of humans to commit errors indicates that, after failures due to human errors (in industry, transportation . . .), adding new prescriptions is probably of little use. A more efficient attitude could be to ask oneself whether or not some situations, tasks or material, due to the nature or human information processing system, would favour human errors. Moreover, it could also be wondered whether some slight modifications of a task would allow operators to correct their errors, since the human brain is equipped of a rather efficient action monitoring system which might allow certain failures to be avoided.

Teachers, may be, should not be that severe as regard errors. Errors sometimes reflect low involvement, negligence or low motivation but, often, they simply reflect the nature of the human information processing system. It seems that errors should, on the contrary, be used for teaching efficient correction and how to avoid them in the future. Finally, an error by itself is often assumed to be a very efficient teacher because learning from past mistakes is essential to ensure future successful behavior (e.g., Holroyd and Coles, 2002). It has been proposed that the Ne reflects a key mechanism for this type of learning from errors (Holroyd and Coles, 2002).

Now, it could be hoped that, in specific situations, if precise enough instructions or long enough teaching is provided, errors could be completely eradicated. Although, from what precedes, one can cast some doubts regarding the results, we wonder whether one should hope complete eradication of errors. Indeed, it is possible that certain general principles of organization are efficient whatever the considered organization level. If this were the case, one could wonder whether some principles of organization of the sensorimotor system could find correspondences in other domains where reliability is also critical. We will try a comparison with the apparently remote domain of DNA replication.

The high (but not perfect) reliability of DNA replication mechanisms is not only dependent of complementary base-pairing.

It also needs different “proofreading” enzymatic mechanisms that sequentially correct mispairing, when present (Alberts et al., 2002).

Complementary base-pairing is not extremely reliable and may produce several initial mispairing. However, certain of these initial mispairing are rendered impossible by DNA polymerase itself which “double-checks” the base-pair configuration before this enzyme catalyzes the covalent binding of mispaired nucleotides. In a second step, if this double-checks mechanism has failed and an incorrectly paired base has been covalently bound to the growing strand, a separate subunit or a separate domain of DNA polymerase, acting as a “self-correcting” system (called “exonucleolytic proofreading”), will correct several of these incorrect pairings. Now, if this self-correcting mechanism also fails, another and last proofreading mechanism (called “strand-directed mismatch repair”) will detect most remaining anomalous pairings and correct them. The end product of DNA replication is, finally, highly reliable and results in a very small proportion of errors (Alberts et al., 2002).

In our terms one could say that the replication process initially engages a preventive (proactive) mechanism with DNA polymerase double-checks. If this proactive mechanism fails, two successive reactive proofreading ones will detect and correct most remaining incorrect base-pairings. [which we could qualify as partial (replication) errors, since covalent binding has already occurred]. Full blown errors are those, when present, which escaped prevention, detection, and correction by the enzymatic replication monitoring system. This enzymatic monitoring system is to be considered as a very sophisticated mechanism which has evolved so efficiently that it powerfully secures the replication process. One might also consider that this system is not completely perfect, yet (since some errors occasionally still occur) and hope that, after a few million more years of evolution, full replication errors will be completely eradicated. However, this should not be: it is clear that maintaining a certain level of errors constitutes a supreme refinement of evolution, without which any adaptation of living beings to changing environments would be impossible. Eradicating completely errors would mean the assured future death of all living species.

If we accept to consider that sensorimotor activities seem to obey similar rules as DNA duplication does, it could also well be that errors should not be eradicated either in sensorimotor activities. Errors might constitute a behavioral avatar of biological variability. This variability might allow exploring new behaviors. Of course, most of these new behaviors reveal to be inappropriate, but some can occasionally reveal to be perfectly fit to an unnoticed change of the environment. In this latter case, these new behaviors, discovered by error, may become the new norm and could even be taught to others. As a consequence, teachers should not be completely negative as regards errors because (1) errors probably contribute to shape efficient and long-living new behaviors, which constitute a sort of self random learning, and (2) errors, even when they do not reveal new appropriate behavior, trigger behavioral adaptations (such as, for example post-error slowing) aimed at avoiding the same kind of errors in the future. This implies that errors, when produced in a given context where they are not too costly, might help avoiding future errors in more costly

contexts. As such, errors might be exploited in a teaching perspective. Therefore, teachers should, to a certain extent, recognize and try to exploit the virtues of errors.

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Perceiving fingers in single-digit arithmetic problems

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In this study, we investigate in children the neural underpinnings of finger representation and finger movement involved in single-digit arithmetic problems. Evidence suggests that finger representation and finger-based strategies play an important role in learning and understanding arithmetic. Because different operations rely on different networks, we compared activation for subtraction and multiplication problems in independently localized finger somatosensory and motor areas and tested whether activation was related to skill. Brain activations from children between 8 and 13 years of age revealed that only subtraction problems significantly activated finger motor areas, suggesting reliance on finger-based strategies. In addition, larger subtraction problems yielded greater somatosensory activation than smaller problems, suggesting a greater reliance on finger representation for larger numerical values. Interestingly, better performance in subtraction problems was associated with lower activation in the finger somatosensory area. Our results support the importance of fine-grained finger representation in arithmetical skill and are the first neurological evidence for a functional role of the somatosensory finger area in proficient arithmetical problem solving, in particular for those problems requiring quantity manipulation. From an educational perspective, these results encourage investigating whether different finger-based strategies facilitate arithmetical understanding and encourage educational practices aiming at integrating finger representation and finger-based strategies as a tool for instilling stronger numerical sense.

Keywords: finger gnosis, arithmetic facts, somatosensory, motor, arithmetic skill

Introduction

Historically and among different cultures, humans have been relying on fingers or body parts to support their representation of numbers (Ifrah, 1994). In occidental cultures, counting on fingers is one of the first strategies taught to children to link the verbal representation of a number with its numerical meaning (Gelman and Gallistel, 1978; Gallistel and Gelman, 1992; Butterworth, 1999; Sato and Lalain, 2008). When counting and calculation procedures are not automatized, fingers alleviate working memory and are important visual cues (Barrouillet and Lépine, 2005; Barrouillet et al., 2008). Children with mathematical learning difficulties rely more on fingers compared to peers either to help represent numerical quantities or to facilitate the execution of operation-specific procedures (Alibali and DiRusso, 1999; Geary, 2005). To date, little is known about the role of finger-based strategies in shaping and creating a strong numerical and arithmetical understanding. Indeed, greater understanding of the neurofunctional changes induced by the use of different finger strategies and their relation to skill will allow implementing better educational practices and hopefully provide alternative tools to remediate mathematical learning difficulties.

Hand and finger representations have been shown to influence children and adults at different levels of numerical processing (Di Luca et al., 2006; Di Luca and Pesenti, 2008; Domahs et al., 2008; Badets and Pesenti, 2010; Badets et al., 2010). Early external finger-based configurations, used to represent numbers and support calculation procedures, become internalized during primary school to the point of influencing adult performance when performing numerical tasks (Di Luca et al., 2006; Di Luca and Pesenti, 2008; Klein et al., 2011). Prototypical finger configurations in adults were responded to faster than atypical ones in an Arabic digit-to-finger mapping task and only these gave an automatic access to number semantics (Di Luca et al., 2006; Di Luca and Pesenti, 2008). Finger-based strategies also influence mental arithmetic (Badets et al., 2010; Klein et al., 2011; Newman and Soylu, 2013). For instance, adults like children show an effect in mental calculation resulting from a failure in keeping track of “full hands” known as the split-five error (i.e., answers to the problems deviated exactly by ± 5 from the correct result; Domahs et al., 2008; Klein et al., 2011). In addition, internalized finger-based representation of arithmetic operations may still rely on motor and somatosensory finger areas in adults since passive finger movement was found to disrupt counting-based strategies during simple mental addition problems (Imbo et al., 2011). Importantly, not only the motor component of finger-based strategies and the mental representation of hand configurations are related to numerical and arithmetical processing, but evidence shows that finger representation *per se*, known as finger gnosis, is related to skill. A lesion in the dominant inferior parietal lobule is found to cause Gerstmann’s syndrome (Gerstmann, 1940) where finger agnosia (i.e., deficit in finger representation) and acalculia (i.e., disability to calculate) are associated suggesting that both competences rely, at least partially, on common processes. The relation between finger gnosis and arithmetic skill has also been found in children independently from IQ scores (Strauss and Werner, 1938; Costa et al., 2011). In 6-year-olds, the quality of the finger representation was a better predictor of mathematical skill than standard developmental tests (Fayol et al., 1998; Noël, 2005; Reeve and Humberstone, 2011) and training in finger discrimination at the same age improved performance to numerical and quantification tasks (Gracia-Bafalluy and Noël, 2008). Interestingly, the development of finger gnosis, non-symbolic numerical abilities and spatial abilities have been found to be correlated independently from age thus further supporting a functional link between the different competences (Chinello et al., 2013).

Both electrophysiological and neuroimaging studies have found evidence for common neural substrates for hand representation and numerical processing. Using transcranial magnetic stimulation (TMS), Sato et al. (2007) reported changes of excitability of hand muscles in participants performing a visual parity judgment task. Similarly, Andres et al. (2007) found changes of excitability in a visual counting task irrespective of the nature of the counting sequence (i.e., numbers or letters) suggesting that hand motor circuits may assist the counting process by keeping track of one-to-one correspondence. In an fMRI study, Kansaku et al. (2007) showed that the left ventral premotor cortex was specifically activated when counting large sequences. In the same study, the crucial role of this area for counting large sequences

(over 20 elements) was confirmed by applying TMS, which disrupted participants’ counting ability. Only one fMRI study independently localized the hand motor representation to investigate number processing related activation (Tschemtscher et al., 2012). Despite the absence of overt hand movements, perceiving numbers as digits or written words enhanced activation in motor and premotor areas contralateral to the preferred hand for counting (i.e., left vs. right hand starters).

Activations in finger-related areas have also been reported during calculation. The left precentral finger area, often combined with fronto-lateral activations, has been found active in several studies (Dehaene, 1996; Rueckert et al., 1996; Pesenti et al., 2000; Stanescu-Cosson et al., 2000; Zago et al., 2001) suggesting that these activations reflect the involvement of a finger-movement network underlying finger counting (Pesenti et al., 2000). The premotor and frontal cortices were also found to be significantly more activated during single-digit additions compared to verbal rehearsal (Hanakawa et al., 2002), were specifically activated for two-digit addition and subtraction problems jointly with bilateral parietal areas (Knops and Willmes, 2014), and were sensitive to difficulty in single-digit multiplication problems (Jost et al., 2009). In a meta-analysis, Arsalidou and Taylor (2011) showed that across fMRI studies, the prefrontal cortex including the precentral gyrus is significantly active in all four basic arithmetic tasks. Indeed, the strong co-occurrence of activation in prefrontal cortex (MFG and premotor cortex) and the IPS has also been highlighted in a recent review on the neurobiological underpinning of mathematical cognition (Ashkenazi et al., 2013).

A strong relation is found between finger representation and arithmetic skill, and neuroimaging studies indicate finger-related activations during calculations but to our knowledge only two studies directly test whether there is overlapping activation of hand representation and arithmetic processing. Using fMRI, the first study showed that simple arithmetic and finger discrimination tasks induce common activations in the horizontal IPS and posterior superior parietal lobule in adult participants (Andres et al., 2012). The relation was found to be stronger for subtraction compared to multiplication problems and for the left compared to the right hemisphere. A second study, focused on the neural networks involved in different numerical tasks (i.e., symbolic and non-symbolic number comparison, symbolic and non-symbolic addition and counting tasks) and their relationship to activations underlying finger representations (i.e., a guided finger movement task) and saccades in children (Krinzinger et al., 2011). The contribution of the finger-related network, including the ventral precentral sulcus, the supplementary motor area and the dorso-lateral prefrontal cortex, was stronger for the addition than for the comparison task.

Importantly, behavioral and neurofunctional data support the existence of operation-specific networks. Behaviorally, adults and children have been shown to rely more on retrieval strategies for multiplication problems compared to subtraction problems, which have been shown to require more on quantity manipulation (LeFevre et al., 1996; Fayol and Thevenot, 2012; Barrouillet and Thevenot, 2013). Indeed, language processes have been found to be more relevant for solving multiplication problems whereas visuo-spatial processes for solving subtraction tasks (Lee and

Kang, 2002; Boets and De Smedt, 2010; De Smedt and Boets, 2010). Neurofunctionally, the brain networks involved in different operations have shown to be partially distinct (Fehr et al., 2007). Multiplication problems have been shown to elicit greater activation within the fronto-temporal network subtending verbal processing whereas subtraction problems have been found to elicit greater activations within the intraparietal sulcus, which is involved in numerical magnitude manipulation (Prado et al., 2011). Importantly, this dissociation has been found to increase developmentally indicating an increased differentiation in the network used to solve each operation (Prado et al., 2014). These results highlight how proficiency in single-digit problems may be achieved through different brain regions supporting different strategies.

Strategies used in school when learning how to solve arithmetic problems could shape the neural networks involved in processing multiplication and subtraction problems. Indeed, children are encouraged to retrieve multiplication problems by using a rote learning approach (Campbell and Xue, 2001) whereas subtraction problems are taught by means of procedures without emphasizing memorization (Dehaene, 1992; Campbell and Xue, 2001; Thevenot and Barrouillet, 2006; Barrouillet et al., 2008). Finger-based strategies are used predominantly to solve subtraction problems (Baroody, 1987; Siegler, 1987), and therefore, differential relation of finger-related activations are expected depending on operation type.

Although evidence suggests a close functional and representational link between fingers and arithmetic, no study has directly investigated how finger-related activation is related to skill in children, or whether these relations depend on operation type. Moreover, we do not know the specific contributions of finger representation (i.e., somatosensory activation) vs. finger movement (i.e., motor activation). Therefore, the aim of the present study was to understand skill-based effects in finger-related activation in both somatosensory and motor cortex during single-digit operations. We specifically used subtraction and multiplication problems because they have shown the greatest dissociation behaviorally and neurofunctionally (LeFevre et al., 1996; Prado et al., 2011, 2014; Fayol and Thevenot, 2012; Barrouillet and Thevenot, 2013). First, we tested whether different operations recruit these areas differently. Subtraction problems require greater quantity manipulation (Dehaene et al., 2003; Barrouillet et al., 2008) and multiplication problems rely more on verbal retrieval (Prado et al., 2011, 2014). In addition, finger movement selectively interfered with addition and subtraction problems but not with multiplication problems (Michaux et al., 2013). Therefore, we expected overall greater involvement of somatosensory and motor areas in subtraction problems. This would be consistent with the greater overlap between numerical processing and finger discrimination found for subtraction than multiplication problems in adults (Andres et al., 2012). Second, we tested the relationship between skill and amount of activation in finger related cortex. Studies on finger gnosis have shown that greater finger discrimination skill was predictive of future arithmetical skill (Noël, 2005; Gracia-Bafalluy and Noël, 2008). We therefore expected that children with better finger somatosensory representations would show higher accuracy. This may be associated with less activation

because a recent study on passive sensory finger stimulation found decreased activity with increased spatial acuity of pinch grip in the somatosensory area (Ladda et al., 2014). Conversely, because children with math difficulties have been found to rely more on finger-based strategies (Alibali and DiRusso, 1999; Geary, 2004, 2005), we expected activation in finger motor areas to be related to lower accuracy.

Materials and Methods

Participants

Forty children (23 females) between 8 and 13 years of age were scanned. One participant was excluded for accuracy in the scanner beyond three SD from the average. Thus, 39 children were retained based on standardized testing performance and fMRI scan quality. All participants had a full-scale IQ standard score greater than 85 on the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) with a group average of 116.3 [SD = 13.8, range (86–144); see Table 1]. To ensure participants had no mathematical difficulty, children had an 85 or above [mean = 106.3, SD = 12.2, range (88–143)] score on the Math Fluency (MF) subtest of the Woodcock–Johnson III (Woodcock et al., 2001). In this task, participants have 3 min to solve one-digit addition, subtraction, and multiplication problems. A timed task was chosen because it is an index of automaticity of procedural strategies and penalizes children that rely on lengthy and immature backup strategies (Russell and Ginsburg, 1984; Fayol and Thevenot, 2012). Finally, all children performed 60% or higher on the fMRI tasks.

Written consent was obtained from children and their legal guardians. The Institutional Review Board at Northwestern University approved all experimental procedures before data collection.

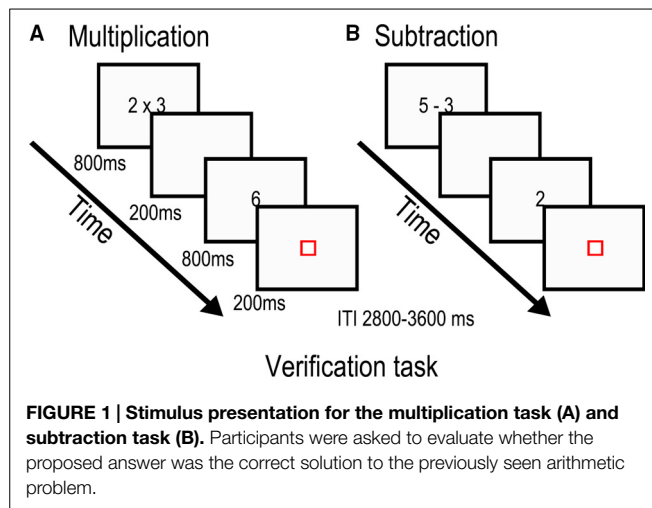
fMRI Subtraction and Multiplication Tasks

Participants performed a subtraction and a multiplication task in the fMRI scanner and were required to judge if a proposed outcome was correct or incorrect responding with the right hand. For each task, based on previous research (Stazyk et al., 1982; Cooney et al., 1988; Siegler, 1988; Ashcraft, 1992; Campbell and Xue, 2001; De Brauwier et al., 2006), twelve small and 12 large one-digit problems were included. Problems involving 0 (e.g., $6 - 0$ or 6×0) or 1 as one of the terms (e.g., $7 - 1$ or 7×1), and ties (e.g., $4 - 4$; $4 - 2$) were excluded from the experiment but were used for practice purposes (i.e., 12 true and 12 false problems). For the subtraction task, problem size was determined by whether the difference between the terms was greater than three (e.g., $5 - 3$

TABLE 1 | Age and standard scores for the 39 participants.

	Average (SD)	Max–Min
Age (year:month)	11 (1:5)	8:2–13:4
WASI-IQ*	116 (13.5)	86–144
WJ-III-Math Fluency*	107 (12.4)	88–143

SD, standard deviation; WASI, Wechsler Abbreviated Scale of Intelligence; WJ-III, Woodcock–Johnson III. *Standard Scores with average 100 (SD = 15).



and $8 - 3$ for small and large problems respectively). For the multiplication task, problem size depended on the size of the operands: small problems had the two operands equal or smaller than 5 (e.g., 2×4) and 12 large problems had both operands larger than 5 (e.g., 6×9). In both tasks, each problem was repeated twice with a true answer and once with a false answer, yielding 72 trials for each problem type (36 small problems and 36 large problems). For the subtraction task, false answers were generated by either adding 1 or 2 (e.g., $6 - 2 = 5$) or subtracting 1 (e.g., $6 - 2 = 3$) from the correct answer. For the multiplication task, the false answer was the correct result of the adjacent fact by adding or subtracting 1 to the first operand (e.g., 20 or 28 as the false answer to 6×4). Twenty-four null trials were included to control for motor responses for each task. In these trials participants had to respond when a blue fixation square turned red.

Stimulus presentation was fixed and identical for the two tasks (Figure 1). The first stimulus was presented for 800 ms before being replaced by a blank screen for 200 ms. The second stimulus was also presented for 800 ms, but was followed by a red fixation square for 200 ms. The red square indicated the need to give a response during a variable interval ranging from 2800 to 3600 ms. Null trials were composed of a blue square that lasted for the same duration as the experimental conditions and participants had to press a button when it turned red. Finally, each run ended with 22 s of passive visual fixation. Each task was subdivided in two approximately 4.3 min blocks to allow for some resting time.

Experimental Protocol

Participants were familiarized with the tasks and the fMRI environment during a practice session after giving informed consent and having completed standardized testing. During this session, they learned to minimize head movement in a mock fMRI scanner by means of an infrared-tracking feedback device and practiced all tasks. This session was completed within a week prior to actual fMRI data acquisition. In the fMRI scanner, each task was split into two 4-min runs. The timing and order of trials within each run were optimized for estimation efficiency using optseq2¹.

Behavioral responses were recorded using an MR-compatible keypad placed in the right hand. Stimuli were generated using E-prime software (Schneider et al., 2002) and projected onto a translucent screen that was viewed through a mirror attached to the head-coil.

fMRI Data Acquisition

Images were collected using a Siemens 3T TIM Trio MRI scanner (Siemens Healthcare, Erlangen, Germany) at CTI, Northwestern University's Center for Translational Imaging. The fMRI blood oxygenation level dependent (BOLD) signal was measured with a susceptibility weighted single-shot echo planar imaging (EPI) sequence. The following parameters were used: TE = 20 ms, flip angle = 80°, matrix size = 128×120 , field of view = 220×206.25 mm, slice thickness = 3 mm (0.48 mm gap), number of slices = 32, TR = 2000 ms. Before functional image acquisition, a high resolution T1 weighted 3D structural image was acquired for each subject (TR = 1570 ms, TE = 3.36 ms, matrix size = 256×256 , field of view = 240 mm, slice thickness = 1 mm, number of slices = 160).

fMRI Preprocessing

Data analysis was performed using SPM8². After discarding the first six images of each run, functional images were corrected for slice acquisition delays, realigned to the first image of the first run and spatially smoothed with a Gaussian filter equal to twice the voxel size ($4 \text{ mm} \times 4 \text{ mm} \times 8 \text{ mm}$ full width and half maximum). Prior to normalizing images with SPM8, we used ArtRepair (Mazaika et al., 2007, 2009)³ to suppress residual fluctuations due to large head motion and to identify volumes with significant artifact and outliers relative to the global mean signal (i.e., 4% from the global mean). Volumes showing rapid scan-to-scan movements of greater than 1.5 mm were excluded via interpolation of the two nearest non-repaired volumes. Interpolated volumes were then partially deweighted when first-level models were calculated on the repaired images (Mazaika et al., 2007). Finally, functional volumes were co-registered with the segmented anatomical image and normalized to the standard T1 Montreal Neurological Institute (MNI) template volume (normalized voxel size, $2 \text{ mm} \times 2 \text{ mm} \times 4 \text{ mm}$). Scan quality was determined by the number of replacements in each functional run: up to 5% of replaced scans, but no more than four consecutive replacements, were accepted for each run.

fMRI Processing

Event-related statistical analysis was performed according to the general linear model. Activation was modeled as epochs with onsets time-locked to the presentation of the first stimulus and with a duration matched to the length of the trial (i.e., 2 s). Trials were classified for problem type (true, false) and for problem size (small, large). However, only true trials were considered of interest in behavioral and fMRI analyses because for false trials it is impossible to determine if the answer was rejected by using a calculation procedure or relying on alternative strategies such

²www.fil.ion.ucl.ac.uk/spm

³http://cibsr.stanford.edu/tools/human-brain-project/artrepair-software.html

¹http://surfer.nmr.harvard.edu/optseq/

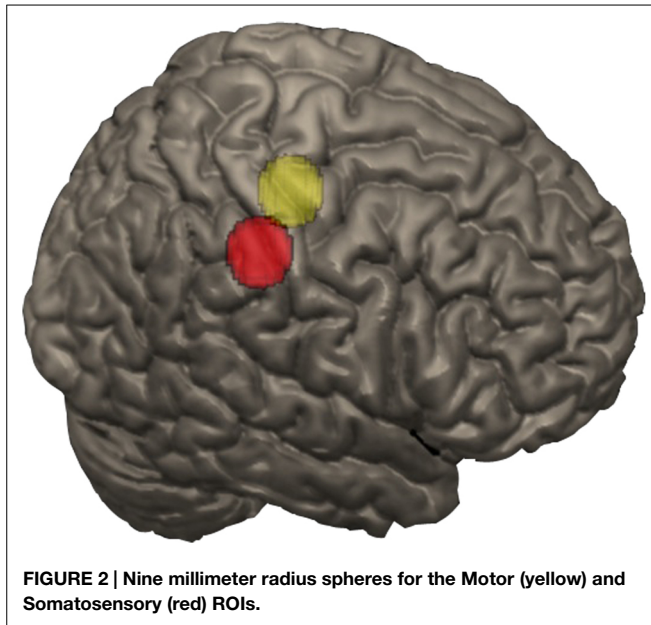


FIGURE 2 | Nine millimeter radius spheres for the Motor (yellow) and Somatosensory (red) ROIs.

as parity judgment or estimation (Lemaire and Reder, 1999). Moreover, during false trials, conflict detection and error monitoring processes could affect activation patterns (van Veen and Carter, 2002, 2006; Ferdinand and Kray, 2014; Ullsperger et al., 2014). Null trials were further modeled in a separate regressor. All epochs were convolved with a canonical hemodynamic response function. The time series data were high-pass filtered (1/128 Hz), and serial correlations were corrected using an autoregressive AR (1) model.

Motor and Somatosensory Region of Interest Definition

To isolate finger-related activation, finger somatosensory and motor areas were defined using Neurosynth, a large-scale automated synthesis of human functional neuroimaging data⁴. Spheres of 9 mm (i.e., 196 voxels) were created by identifying the peaks in the pre- and postcentral gyrus forward inference maps for fingers as centers. Only the right hemisphere was considered since participants were using their right hand to give their response and the left hand was available as support for calculations. The somatosensory peak was at 46, -30, 44 and the motor peak was at 38, -22, 60 in MNI coordinates (Figure 2).

fMRI Analyses

Voxel-wise regressions were run within the two region of interests (ROIs) to investigate the relation with accuracy. Age was entered as control variable and accuracy for each task or condition as variable of interest. An uncorrected height threshold was set at $p < 0.01$ for the contrast maps and Monte Carlo simulations (3dClustSim) were used to set the extent threshold at $p < 0.05$ corrected. Statistical results are reported in the MNI coordinate space.

⁴www.neurosynth.org

TABLE 2 | Accuracy and reaction times (RTs) for the subtraction and multiplication tasks.

Task	Accuracy (SD)	RT (SD)
Subtraction	91 (8.9)	1173 (336)
Small problems	93.7 (6.6)	1058 (318)
Large problems	88.3 (12.4)	1252 (380)
Multiplication	88 (10.5)	1120 (358)
Small problems	96.7 (5.2)	960 (329)
Large problems	79 (18.4)	1239 (380)

SD, standard deviation.

Results

Behavioral

Repeated measures ANOVAs were run with operation and problem size as within subject variables on accuracy and reaction times (RTs). Large problems were significantly slower [$F_{(1,38)} = 78$, $p < 0.001$] and less accurate [$F_{(1,38)} = 51$, $p < 0.001$] than small problems (Table 2). Moreover, for accuracies only, the interaction indicated that the problem size effect was larger for multiplication problems [$F_{(1,38)} = 19$, $p < 0.001$]. All other effects were non-significant.

Age was significantly correlated with subtraction accuracy ($r = 0.403$, $p = 0.011$) and only marginally to multiplication accuracy ($r = 0.309$, $p = 0.056$), but not with RTs. Controlling for age, accuracies to the two tasks were also correlated ($r = 0.428$, $p = 0.007$).

Operation Related Activation

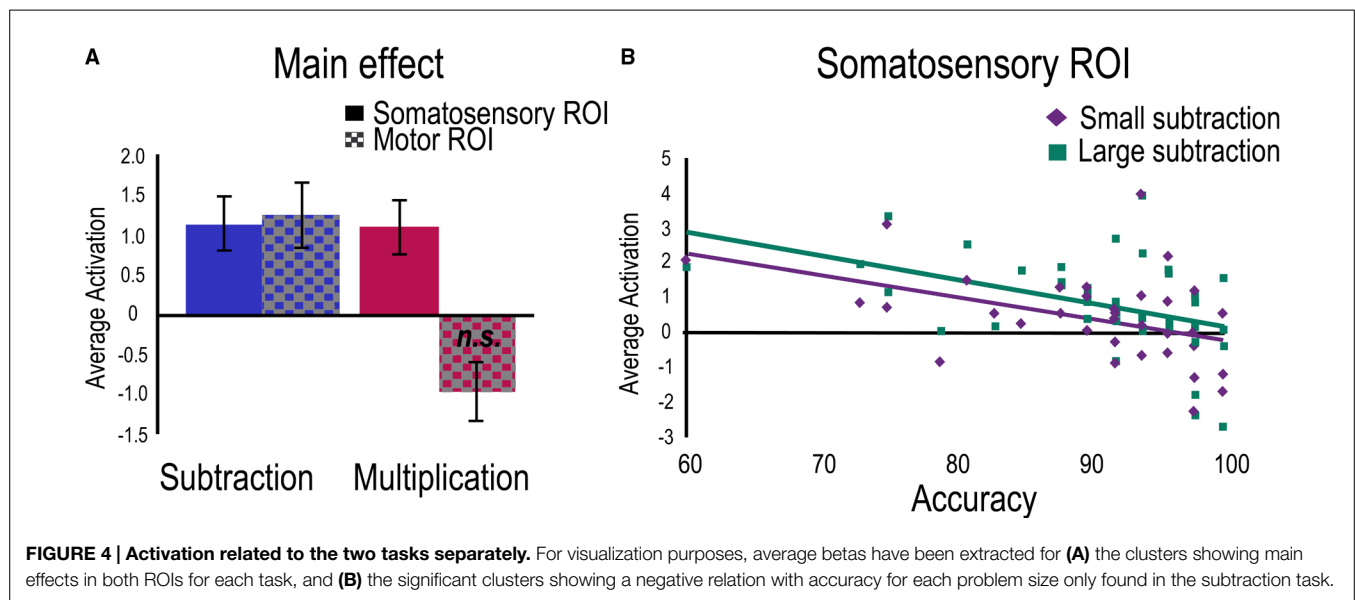
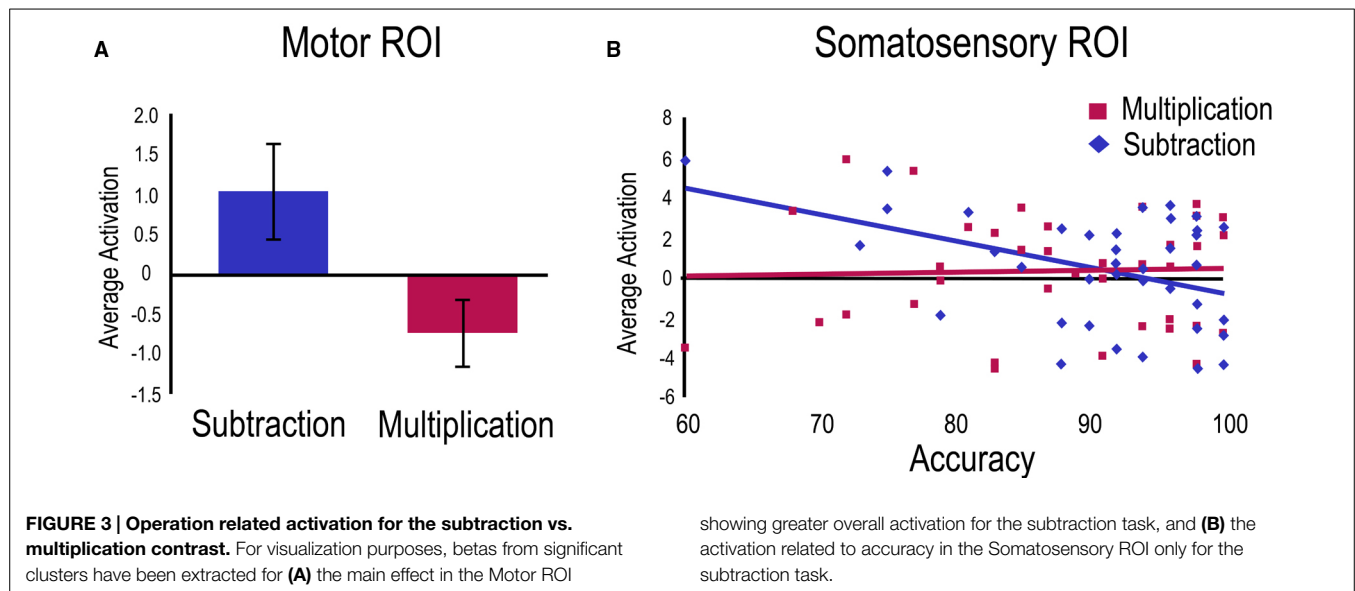
A first regression was run on the subtraction vs. multiplication contrast with age as control variable and accuracy as a predictor. A main effect of task was found within the Motor ROI indicating greater activation for subtraction problems compared to multiplication problems (Figure 3A). Within the Somatosensory ROI, a negative relation was significant with subtraction accuracy (Figure 3B).

Multiplication Task

Separate regressions were then run for each task with age as control variable and accuracy as a predictor. For the multiplication task, a positive main effect was found within the Somatosensory ROI and a marginally negative main effect in the Motor ROI (Figure 4A). No relation with accuracy was found significant. To investigate problem size, a regression for the large vs. small contrast was run with age as control variable and accuracies to small and large problems as predictors. No main effect or relations with accuracies were found significant.

Subtraction Task

For the subtraction task, significant positive main effects were found in both ROIs (Figure 4A) and the negative relation between accuracy and activation was confirmed within the Somatosensory ROI. A regression to investigate problem size (large vs. small contrast) was therefore run with age as control variable and accuracies to small and large subtraction problems as predictors. A main effect was found in the Somatosensory ROI indicating greater



engagement for large problems compared to small problems. Separate regressions were also run for large and small problems separately. For large problems, positive main effects were found in both ROIs and a significant negative relation was found with accuracy only in the Somatosensory ROI. For small problems, only the negative relation with accuracy was significant (**Figure 4B**).

Conclusion

Evidence suggests that finger representation and finger-based strategies play an important role in learning and understanding arithmetic (Alibali and DiRusso, 1999; Geary, 2005; Gracia-Bafalluy and Noël, 2008; Newman and Soylu, 2013). However, no study has investigated in children the neural underpinnings of finger representation and finger movement involved in arithmetic

and their relation to skill. This is the first study to specifically investigate skill based effects in finger-related areas in somatosensory and motor cortex during single-digit problems. Previous studies have shown that different operations rely on different processes (Arsalidou and Taylor, 2011; Ashkenazi et al., 2013) and that interference from finger movements depends by operation type (Michaux et al., 2013); therefore we also tested whether hand-related activation varied depending on the operation. We compared activation for subtraction and multiplication problems in independently localized finger somatosensory and motor areas and tested whether activation was related to skill.

Comparing activations for the two operations, we found that children recruited motor areas more for subtraction problems suggesting that they were more likely to support their mental processes with finger-based back up strategies. This is consistent

with evidence suggesting that subtraction problems require more quantity manipulation (Dehaene et al., 2003; Barrouillet et al., 2008) and multiplication problems rely more on verbal retrieval (Prado et al., 2011, 2014). Against our initial hypothesis, we did not find finger motor activation to be related to skill. Because previous behavioral studies have found that children with math difficulty rely more on finger-based strategies than typically performing peers (Alibali and DiRusso, 1999; Geary, 2004, 2005), we expected to find a relation between motor activation and skill. Additionally, we showed no difference in motor activation with problem size. A possible explanation is that children were relying on some finger-based strategy irrespective of problem size but the use of this strategy did not yield better performance. Conversely, because children were discouraged to move in the scanner, it could be that they were inhibiting explicit finger movements but still showed activation in the motor area. Consistent with the latter explanation is the study from Tschentscher et al. (2012). The authors report activation in the motor and premotor areas contralateral to the preferred hand for counting when perceiving numbers, digits or written words, despite the absence of overt hand movement. Finger-based and counting strategies may become internalized to the point of influencing adult performance during numerical tasks (Di Luca et al., 2006; Di Luca and Pesenti, 2008; Klein et al., 2011). Our result suggests that the observed influence might be the consequence of an implicit activation of the areas supporting these finger-based strategies.

In both tasks, we found significant activation in the somatosensory area suggesting an implicit activation of finger representation. Indeed, several studies have shown that finger representation is automatically recruited when processing numerical information (Di Luca et al., 2006; Di Luca and Pesenti, 2008; Badets and Pesenti, 2010; Badets et al., 2010). A critical finding of our study is that activation in the somatosensory area was related to performance only for subtraction problems. Studies have shown that finger gnosis, that is the quality of an individual's finger representation, in children was related to arithmetical skill and training finger representation also improved performance to arithmetic tasks (Fayol et al., 1998; Noël, 2005; Gracia-Bafalluy and Noël, 2008; Reeve and Humberstone, 2011). Our result suggests that finger representation has greater functional involvement for operations requiring greater quantity manipulation. Importantly, the relation indicated that children with low performance engaged somatosensory areas more than children with higher performance. A first possible explanation is that low performers relied more on an immature finger-based strategy whereas high performers possibly retrieved the answer. However, this explanation seems unlikely because it should also reflect in a negative relation within the motor area, but this was not found. The second and more likely explanation is that activation in the somatosensory areas is negatively related with the quality of fingers representation: participants with greater finger gnosis have lower levels of activation within the somatosensory area. In support, studies on sensory finger stimulation showed decreased activity within the somatosensory area after training along with increased spatial acuity (Ladda et al., 2014). Therefore, the negative relation may indicate that children that performed better on subtraction problems were also those with finer finger representation.

We also found that the engagement of the somatosensory cortex was modulated by problem size only within subtraction problems. Large problems engaged the somatosensory area more than small problems. We suggest that more fingers are needed to represent large numbers thus requiring greater engagement of the somatosensory area. It can be argued that large problems might be harder and induce more finger-based backup strategies. However, these problems did not engage more motor activation compared to small problems suggesting that children were not more likely to rely on finger-based strategies. This is in contrast to previous studies that have shown greater activation in motor related areas for arithmetic tasks. In a study using single-digit multiplication problems, along with activity found in retrieval areas, the authors also found increased activity for harder problems in premotor and frontal cortices (Jost et al., 2009). A network including the IPS, the inferior frontal gyrus and the precentral gyrus also showed increased activation for larger single-digit addition problems (Stanescu-Cosson et al., 2000). It can be hypothesized that larger problems required greater computation thus involving more motor hand support. The differences between our study and previous ones could be ascribed to task presentation differences. In Jost et al. (2009) participants had to produce the answer to consecutive multiplication and addition problems. This paradigm might have increased working memory load thus increased the reliance on finger-based strategies. In Stanescu-Cosson et al. (2000) study, additions problems were presented in blocks divided by problem size. This presentation format might have induced different strategies depending on problem size due to differences in cognitive load between the blocks. Because large problems are also usually harder, participants might have more often used back-up strategies for such blocks. In our study, children had to judge the accuracy of small and large intermixed problems. This presentation might have decreased the chances of using different solutions depending on problem size.

Two alternative explanations for these results need to be discussed. First, studies have shown that the visual presentation of symbols activates a complex network including sensorimotor, premotor and motor areas corresponding to the graphic movements involved in tracing or writing (Longcamp et al., 2003, 2005; James and Gauthier, 2006). However, because the stimuli presented were single-digits in both tasks, it is unlikely that the differences in motor and sensorimotor activations found in our study can be ascribed to differences in the network subtending such movements. It is unlikely that only the symbols for the subtraction task elicited a graphic motor network. Our results also show a relation between performance and activation in the somatosensory area, which is not consistent with graphic-related motor activations. Additionally, Exner's area, known as the "graphic motor image center" in the frontal lobe, is situated more anteriorly compared to our ROIs (Roux et al., 2009; Purcell et al., 2011, for a meta analysis). Second, a limitation of this study is the use of a manual response. Indeed, studies have shown that motor responses induce activations both in the contralateral and ipsilateral hemisphere, which could interact with our results (Buettefisch et al., 2014). However, previous studies also show that activation found in the ipsilateral hemisphere is spatially distinct

from activation found during the contralateral task (Cramer et al., 1999). If contralateral activation is modulated by task difficulty, where greater cognitive load increases ipsilateral motor activation (Buetefisch et al., 2014), our brain-behavior results should have shown a relation with multiplication problems rather than subtraction problems because they show a larger problem size effect. Additionally, because the two tasks were well matched in overall accuracies, it is unlikely that the differences observed between tasks were induced by greater ipsilateral activation for correct responses only in the subtraction task. Finally, the amount of ipsilateral motor button response cannot explain differences in activation because in both tasks participants required a response for each trial (i.e., true or false).

Overall, our results support the importance of fine-grained finger representation (i.e., finger gnosis) in performing subtraction problems and are the first evidence for a functional role of the somatosensory finger area in proficient arithmetical problem solving, in particular for those problems requiring quantity manipulation. Although training studies show a causal role of finger gnosis in arithmetical skill (Gracia-Bafalluy and Noël, 2008), future studies should investigate the differential impact depending on operation. Our results suggest that training should have more pronounced effects on subtraction compared to multiplication. Studies should also investigate the neurofunctional changes associated with finger training and their relation to performance. It is possible that training may enhance the relation between amount of activation and accuracy in somatosensory cortex, or alternatively, it could weaken the relationship due to greater benefit for low skill participants. Finally, because children with difficulties in learning mathematics show lower finger gnosis (Strauss and Werner, 1938; Noël, 2005; Costa et al., 2011), it would be interesting to investigate whether these children differ in their ability to recruit somatosensory areas during arithmetical problem solving and whether it relates to performance. A logical extension of our results would predict that children with mathematical learning disability would show even greater recruitment of somatosensory cortex during arithmetic problems.

Currently, educational practices have considered finger counting as a tool to introduce the transformations associated to each operation and as an initial support to alleviate working memory

(Alibali and DiRusso, 1999; Geary, 2005). However, little attention has been given to the types of counting strategies and their implications on performance. Great cultural variability has been described and different types of finger counting strategies possess different properties (Bender and Beller, 2012). It is therefore important to understand developmentally if different properties such as dimensionality, base and sub-base values, regularity or number of distinct finger configurations might prove more efficient in fostering understanding of numerical operations. For example, some studies suggest that the spatial-numerical association, which in turn influences arithmetical processing (Knops et al., 2009, 2014), might be influenced by finger counting strategies. Given our results showing motor and somatosensory activations dependent on operation type, systematically studying the influence of different finger strategies might be a promising area of research with direct educational implications.

In conclusion, these results support educational practices encouraging the use of fingers as functional link between numerical quantities and their symbolic representation as well as an external support for learning arithmetic problems. These results also encourage educational practices to focus on finger discrimination as a precursor of numerical and arithmetical skill. Finally, although the National Mathematics Advisory Panel (2008) suggests that children should achieve automatic retrieval for all arithmetic facts, regardless of the operation, our study strengthens the hypothesis that different arithmetic operations are processed in distinct ways and successful performance might be achieved through at least partially different neurofunctional processes.

Author Contributions

IB collected, analyzed and interpreted the data, conceived the work and wrote the manuscript; JRB conceived the work, interpreted the data, and critically revised the work.

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Basic and supplementary sensory feedback in handwriting

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The mastering of handwriting is so essential in our society that it is important to try to find new methods for facilitating its learning and rehabilitation. The ability to control the graphic movements clearly impacts on the quality of the writing. This control allows both the programming of letter formation before movement execution and the online adjustments during execution, thanks to diverse sensory feedback (FB). New technologies improve existing techniques or enable new methods to supply the writer with real-time computer-assisted FB. The possibilities are numerous and various. Therefore, two main questions arise: (1) What aspect of the movement is concerned and (2) How can we best inform the writer to help them correct their handwriting? In a first step, we report studies on FB naturally used by the writer. The purpose is to determine which information is carried by each sensory modality, how it is used in handwriting control and how this control changes with practice and learning. In a second step, we report studies on supplementary FB provided to the writer to help them to better control and learn how to write. We suggest that, depending on their contents, certain sensory modalities will be more appropriate than others to assist handwriting motor control. We emphasize particularly the relevance of auditory modality as online supplementary FB on handwriting movements. Using real-time supplementary FB to assist in the handwriting process is probably destined for a brilliant future with the growing availability and rapid development of tablets.

Keywords: handwriting, sensory feedback, vision, proprioception, audition, sonification, enriched reality

INTRODUCTION

Handwriting is described as a complex perceptual-motor skill encompassing a blend of visual-motor coordination abilities, motor planning, cognitive, and perceptual skills, as well as tactile and kinesthetic sensitivities (Feder and Majnemer, 2007). Thousands of hours of practice are required to master handwriting skills. Between 12 and 30% of children fail in the motor learning of handwriting (Rubin and Henderson, 1982; Hamstra-Bletz and Blöte, 1993; Smits-Engelsman et al., 2001; Karlsdottir and Stefansson, 2002). These children are considered as poor writers or as having a dysgraphia, namely a learning disability that concerns the mechanical handwriting skill, unrelated to reading or spelling abilities (Hamstra-Bletz and Blöte, 1993). The ability to control graphic movements clearly impacts on the quantity and quality of the written text (Jones and Christensen, 1999). Handwriting control is based on an efficient treatment of feedback (FB). FB is considered here as sensory information that arises from movement (Schmidt and Lee, 2005). Not properly processing the FB generated by handwriting movements could result in poor handwriting and hence impact the academic success of the child.

Since the early 1980s, many studies have investigated the role of sensory FB in the motor control of handwriting (e.g., Smyth and Silvers, 1987; van Galen, 1991; Teasdale et al., 1993; van Galen et al., 1994; Teulings, 1996). Even if no striking change has recently occurred in the way we write that would justify questioning again the sensory signals involved in the perception and control of handwriting movements, the tools we now have at our disposal to study handwriting have dramatically changed and the

importance of FB can be reconsidered. Thanks to graphic tablets, we are now able to analyze and closely follow the handwriting process, i.e., the movement generating the trace. Consequently, we can analyze and “dissect” handwriting as a movement *per se*, and not indirectly from the static written trace resulting from this movement. Beyond analysis, it has also become possible to act in real-time on this movement in order to change its control. New technologies have improved existing techniques or enabled new methods of supplying the writer with real-time computer-assisted FB. The possibilities are numerous and various. Therefore, two main questions arise: (1) What aspect of the movement is concerned and (2) How can we best inform the writer to help them to correct their handwriting? These two points, the FB contents and the sensory modality involved, all have to be considered together. Depending on their contents, some sensory media will be more appropriate than others to assist handwriting motor control.

The aim of the present review is to make a synthesis of studies devoted to real-time sensory FB in handwriting in order to evaluate the effectiveness of experimental attempts to improve its learning and rehabilitation. We aim also at envisaging new possibilities. For that, we will firstly report studies on FB naturally used by the writer to control his handwriting. The purpose is to determine which information is naturally carried by each sensory modality, how it is used in handwriting control and how this control changes with practice and learning. Secondly, we will report studies on supplementary FB provided to the writer to help him/her to better control and learn how to write. We will discuss the relevance of each sensory modality according to the information content.

BASIC SENSORY FEEDBACK IN THE CONTROL OF HANDWRITING

Considering motor skills in general, two modes of control are classically distinguished: a *proactive* control, based on memorized information defined as *internal model* (Wolpert et al., 1995; Wolpert et al., 2011) or *motor program* (van Galen et al., 1994; Schmidt and Lee, 2005) and a *retroactive* control based on sensory FB. Proactive refers to the components of the movement that are anticipated and prepared before the movement is triggered. All these elements are somehow included in the motor command, the role of FB being just to confirm that everything occurs as it was foreseen. Conversely, retroactive refers to all the aspects of the movement which are not programmed before the onset of the movement and which have to be controlled during the ongoing movement on the basis of sensory FB. In addition, sensory FB is also used during learning to improve the planning of the following movement and hence the proactive component. Skilled handwriting in particular involves both types of control: It cannot be totally proactive since several aspects of its production should be controlled in-line. It is considered as semi-automatic. During handwriting learning, Meulenbroek and van Galen (1986, 1988) observed a switch from a proactive (around 5 years) to a retroactive control (around 7 years), followed by a mixed control in older children (around 10 years) when handwriting mastering and motor maturity are reached.

Two types of sensory FB, visual and proprioceptive, are naturally used in handwriting. Two questions arise about them: (1) What are their respective roles and interactions, and (2) How do their roles and interactions change during learning?

VISUAL FEEDBACK

Visual FB informs about the spatial characteristics of the written trace: where and how the trace is produced. To try to understand the role of vision, many authors have studied the changes of handwriting caused by the absence or the deterioration of visual FB (see Table 1).

Two types of constraints have to be dealt with in handwriting production: those related to the formation of the letter's shape and those related to the spatial layout and sequencing of the letters on the paper. Paillard (1990) referred to these two types of spatial constraints as the 'morphocinetic' and the 'topocinetic,' respectively. The morphocinetic component of handwriting concerns the shape of the letters (the "what?" in handwriting). In skilled handwriting, the shape of the letters is perfectly known and mastered; therefore the morphocinetic component is almost independent of visual control. Alternatively, the topocinetic component, which concerns the spatial layout of the text in the graphic space, the spacing between letters and words, the placement of punctuation, etc. (the "where?" in handwriting) requires visual FB. The two components are not identically controlled by visual FB: in the case of experts, suppressing visual FB mainly affects the topocinetic component, without compromising the morphocinetic one (Paillard, 1990). Nevertheless, when letters are complex and composed of several strokes, they are produced under visual control. As a matter of fact, a decrease in the number of strokes and an alteration of the sequence and direction of movements has been shown when writers did not use visual

information (Smyth and Silvers, 1987; Smyth, 1989). Visual FB would update information concerning letters with repetitive strokes in the motor buffer memory (van Galen et al., 1989; van Galen, 1991). In the same vein, Tamada (1995) investigated the effects of delayed visual FB on the handwriting of some familiar words, using various delays between 0 and 500 ms. She observed that writing errors increased with the delay, with a tendency for some additional strokes to be inserted, especially for letters with repetitive strokes, which is in accordance with the model of van Galen et al. (1989).

The handwriting perturbations induced by the absence of vision revealed the crucial role vision plays in the control of the written trace. Nevertheless, expert writers are able to minimize the impact of vision suppression by developing adaptive strategies. This conclusion was drawn by van Doorn and Keuss (1992) by evaluating the movement time and the reaction time in handwriting both with and without vision. They showed that preserving the morphocinetic component required more time when unseen. They confirmed different strategies in the handwriting production without vision to maintain, to a certain level, a spatial invariance (van Doorn and Keuss, 1993). According to another hypothesis, increasing proprioceptive FB during execution could compensated for the absence of vision. This hypothesis was strengthened by the observation that an increase of pressure and size occurred in the absence of vision, as if the writers tried to maximize their proprioceptive perception (van Doorn, 1992).

Consequently, there is general agreement that writing without vision changes the written trace, particularly the topocinetic components. But, does it also affect the kinematics of the handwriting movement? Marquardt et al. (1996, 1999) analyzed the velocity profile both with and without visual FB. When writing both with and without vision, all subjects produced a smooth and single-peak velocity profile. In a second experiment, the authors manipulated the visual control of the written trace, at normal size, 133 and 66% of the normal size. They confirmed that visual FB is not required to produce and control automated handwriting movement but rather to take into account the spatial constraints of the trace (e.g., the size etc.), which seems to hamper the elicitation of automated movement, a conclusion already drawn by Burton et al. (1990).

In conclusion, vision plays a crucial role in the control of the written trace and allows the writer to correctly link words, letters, and strokes within letters (those with repetitive strokes). However, the absence of vision does not significantly affect the handwriting process, i.e., the ongoing movement that generates the trace. Quite the reverse, suppressing vision while writing would promote a more proactive control that would help the "blind" writer to write fluently, at the risk of being less accurate and, consequently, less legible.

PROPRIOCEPTIVE FEEDBACK

Proprioceptive FB arises from muscles, tendons and joints receptors and informs about the positions and movements of limbs. In principle, tactile perception conveyed by cutaneous receptors is not directly included in proprioception. However, for the sake of simplicity we will not consider it separately from proprioception, particularly because when deafferented patients lose

Table 1 | Experimental studies on the role of vision in skilled handwriting reported in a chronological order.

Authors	Number of participants	Relevant comparisons	Experimental task	Data analysis	Main results and discussion
Smyth and Silvers (1987)	12 adults	V vs. VC vs. VB vs. NV vs. NVC vs. NVB	Writing of eight sentences	Writing time; Orientation of writing; Writing errors	The orientation of the written words was affected by the loss of vision rather than by the addition of a secondary task. The writing errors were very similar for the secondary task condition and the without-vision condition.
van Galen et al. (1989)	10 adults	V vs. NV	Writing a list of words, with similar vs. dissimilar upstroke, with and without stroke repetitions within letters, and with long and short letters	MT of correct words	The deprivation of visual FB slowed down handwriting, especially in combination with the conditions that affect the short term memory.
Smyth (1989)	12 adults	V vs. NV	Writing nine letters, nine reverse letters, and drawing eight shapes	Number of strokes; Percentage of occasions on which two start and three progression rules were obeyed in visual and non-visual conditions; Writing errors	Without vision, movement production is simplified to reduce the number of relocations required. The use of consistent directions of movement depends of the ability to use visual control of spatial location.
Burton et al. (1990)	8 adults	V vs. NV in one-fourth, one-half, double, and four-times their normal size	Writing the words 'poppy' and 'wood'	Width of each word; width of the space between the words; width of spaces between letters; width of individual letters; height of 'tall' letters (p, y, and d)	Size transformations were greater and closer to the instructed values with than without vision. Variability was greater with than without vision for all three space segments, with no significant effect of vision for any of the three letter segments. With vision, subjects differentiated letters and spaces in making their horizontal transformations; without vision, there was no differentiation.
van Doorn and Keuss (1992)	12 adults	V vs. NV, in the normal vs. short durations	Writing the sequence 'lenehele'	RT; MT; Size; Spatial variability of the sequence; Variability of MT and Size	Spatial control was not affected by absence of vision but RT increased without vision, suggesting that invariance of shapes is preserved in the absence of vision at the expense of processing time increments.
van Doorn and Keuss (1993)	12 adults	V vs. NV	Writing the sequence 'lelele'	Size; X/Y -ratio; Spatial variability	Geometric aspects of letters altered under no vision and under the scaling requirement to write in a small format.

(Continued)

Table 1 | Continued

Authors	Number of participants	Relevant comparisons	Experimental task	Data analysis	Main results and discussion
Tamada (1995)	8 adults	Visual FB with various delays – 0, 33, 67, 100, 133, 167, 267, and 500 ms	Writing a word	Writing error rate	With increasing the delay, the writing error rate increased, especially in stroke repetition within words. (e.g., “feeling” as “feeling”). Visual monitoring would be indispensable in producing stroke repetition.
Marquardt et al. (1996)	31 adults	Experiment 1: V vs. VT vs. NV vs. MT Experiment 2: VWT in normal size, 133 and 66% of the normal size	Experiment 1: writing the sentence ‘Die Hunde bellen laut’ Experiment 2: writing ‘ll’ repeatedly for 8 s	Number of Inversion in Velocity per stroke; Size	Vision is not required to produce automated handwriting movements and conscious attention to visual control hampers the elicitation of automated movements. Vision would be used to monitor script size even in highly automated handwriting.

V, writing with vision; VC, with vision while counting; VB, with vision while saying ‘blah’; NV, writing with no vision; NVC, with no vision and counting; NVB, no vision and ‘blah’; VT, visual tracking of the pen; MT, mental tracking; VWT, vision of the written trace only; MT, movement time; RT, reaction time.

proprioception, they also lose tactile perception. Kinesthesia refers to sensory information about movements, not including static positions: therefore, kinesthesia and proprioception do not totally overlap. In handwriting, kinesthetic FB can inform about spatial, kinematic, and/or dynamic characteristics of handwriting movement whereas tactile FB from skin receptors can inform about the pressure exerted by the fingers onto the pen and thus about the forces exerted during handwriting. Of course, removing proprioception is not as easy as closing the eyes, therefore quantifying how far a writer takes into account proprioceptive FB is difficult. This is why only a few studies have been conducted and they only investigated the absence of proprioceptive information in study-cases of deafferented patients (e.g., Ghez et al., 1990; Teasdale et al., 1993; Hepp-Reymond et al., 2009).

Teasdale et al. (1993) asked a deafferented patient to write with and without vision. The comparison of the trace written by the patient using vision and that of the control subjects did not reveal a difference attributable to the lack of proprioception. However, when the patient’s handwriting with and without vision was compared, only the topocinetic component was affected by the absence of vision, i.e., when she was lacking all FB. In other words, the absence of proprioceptive FB did not affect the written trace: The morphocinetic component (form of the letters) was preserved and only the topocinetic component deteriorated due to the absence of both vision and proprioception. These findings confirm that, in skilled handwriting, proprioceptive FB is not fundamental for controlling either the shape of the letters or their spatial layout on the page: the former would be controlled by a proactive mode and the latter by visual FB.

If proprioception does not inform about the “product” of handwriting (the written trace), does it inform about the process (the movement)? To try to answer this question, Hepp-Reymond et al. (2009) quantified precisely the role of proprioception and vision in a deafferented patient and healthy participants. They compared 13 handwriting variables in the cursive writing of a word. Where Teasdale et al. (1993) concluded that proprioception played a weak role in handwriting trace, Hepp-Reymond et al. (2009) showed that handwriting movement was clearly affected by the lack of proprioception. More precisely, they demonstrated that three variables (number of pen lifts, number of inversions in velocity profile, and mean stroke frequency) changed without proprioceptive FB, whatever the visual conditions. In the patient who lacked proprioception, the written words remained legible provided she benefited from visual FB, but the movement was affected.

Whether the movement deterioration only results from a lack of kinesthetic FB, from a lack of tactile FB or from both, remains an open question. Only one study (Ebied et al., 2004) was devoted to the lack of tactile FB in healthy participants (by infiltrating a local anesthetic around the median nerve at the wrist). The authors observed that blocking cutaneous sensation did impair the ability to write, as judged by an increase in the movement time and in acceleration fluctuations. These findings highlight the importance of touch in handwriting control, though more studies are necessary to provide clear conclusions about its specific role. The number of studies devoted to the role of cutaneous FB is probably going to increase in the

future, with the rapid development of smartphones and tactile tablets in which the pen tends to be replaced by the finger (e.g., Tu and Ren, 2013).

In conclusion, proprioceptive FB does not really seem to inform about the spatial characteristics of the written trace, but it is useful for controlling the kinematics and dynamics of handwriting movement. Therefore, suppressing proprioceptive FB has the opposite effect of suppressing visual FB, confirming that vision and proprioception are clearly complementary in handwriting control.

What does happen when visual and proprioceptive FB are not congruent? Changing the congruence of the visual FB, for instance in a mirror-drawing task, induces a conflict between visual and proprioceptive FB. This conflict permits the study of the relative contributions of the two sensory modalities. Lajoie et al. (1992) demonstrated that a deafferented patient had no problem achieving a mirror-drawing task, whereas healthy participants needed more than four trials to attain a similar performance. They proposed that the inversion of visual coordinates imposes a recalibration because of the conflict with proprioceptive FB. In the deafferented patient, the conflict does not exist. Using the same mirror-drawing protocol in healthy subjects, Gullaud-Toussaint and Vinter (1996, 2003) observed the dominance of either the visual or the proprioceptive modality within the visual-proprioceptive conflict. They distinguished two strategies, one favoring vision which preserved the perceived movement directions, but in turn induced a reversal of the directions drawn on the sheet of paper, the other favoring biomechanical constraints which tended to preserve the directions drawn on the sheet of paper, but in turn provoked a reversal of the directions perceived in the mirror condition.

HOW DO VISUAL AND PROPRIOCEPTIVE FEEDBACK CHANGE WITH LEARNING AND DEVELOPMENT?

Mastering handwriting requires several years of practice usually achieved during childhood, therefore, handwriting motor control, and hence the use of FB, change both with the increase in learning and development of the brain and body (e.g., Chiappedi et al., 2012). Handwriting movement control evolves from a retroactive to a more proactive mode with learning (Schmidt and Lee, 2005). However, the change in handwriting control is not monotonic during the child's development: it evolves from an initial predominance of fast ballistic movements at 5–6 years to the mature medium-speed ballistic movements, around 9–10 years, via a relatively unstable period at 7–8 years (Meulenbroek and van Galen, 1986, 1988).

Meulenbroek and van Galen (1986, 1988) interpreted these changes as a switch from a proactive (around 5 years) to a retroactive control (around 7 years), followed by a mixed control in older children (around 10 years). Asking 6 to 9 year-old children to increase their execution speed improved the fluency. Since increasing the speed reduces the time available to take into account the FB, they concluded that it would induce a more proactive mode of control. In the same vein, Chartrel and Vinter (2008) studied the effect of temporal (speed) and spatial (size) constraints on cursive letter production in 5 to 7 year-old children. The idea was that temporal constraints decrease visual control and hence improve fluency. They observed the same effect of spatio-temporal constraint at the

age of 6 and 7 years, but not at 5 years. In line with the study by Hay (1984), they suggested that 5 year-old children control their movement more proactively. In conclusion, additional spatial cues may help visual FB in young children and speed constraint would be more beneficial to older children.

In addition to a global reduction of FB use, increasing expertise in motor control can be explained by a gradual change in the balance between visual and kinesthetic control (Fleishman and Rich, 1963; Schmidt and Lee, 2005). The underlying argument was that at the beginning of learning, the learners do not have a kinesthetic reference of the movement and hence control it visually. With practice, this kinesthetic information is memorized and then used as reference for executing the following movements, thus reducing the need for visual control. Laszlo and Bairstow (1984) aimed at linking handwriting performance in 5 to 6 year-old children with kinesthetic sensibility. They showed that children who, following specific training, had improved their kinesthetic sensitivity, had also improved their handwriting skills. They concluded that the lack of kinesthetic readiness, a term proposed by these authors, explains the difficulty that may hinder effective training of writing at this age. They suggested delaying formal training of handwriting until the age of seven, when most children develop kinesthetic readiness naturally. However, examining the effect of kinesthetic training on the handwriting performance in first graders, Sudsawad et al. (2002) did not successfully link kinesthesia and handwriting. The positive effect of kinematic training on handwriting remains unclear and should be considered with caution.

Does visual FB have the same importance in children who are learning to write and in adults mastering their handwriting? We previously mentioned that only the spatial organization of the written trace was affected by the absence of visual FB in adults: not the movement kinematics. Chartrel and Vinter (2006) compared the role of visual FB in 8 to 10 year-old children and they showed that the absence of visual FB was more detrimental in younger (8 year-old) than in older (10 year-old) children and adults. Without vision, movement time increased and movement fluency decreased in the youngest children whereas the handwriting kinematics was not changed by absence of visual FB in adults. To conclude, visual FB would be crucial in children who are beginning to learn how to write and who have not yet a complete representation of the shape of letters. In addition, the younger children may not be able to process the proprioceptive signals about movement dynamics. Therefore, suppressing the visual FB in children would probably lead them to write differently, for instance at a larger size, resorting more to proprioceptive signals, but without favoring the automatization of movement.

CONCLUSION

Two sensory modalities are naturally used for controlling handwriting: vision and proprioception. Visual FB is used mainly for controlling the spatial layout of the written trace. Proprioceptive FB is used for controlling movement execution, thus freeing vision for other controls. The lack of proprioceptive FB does not affect the capability of writing a legible trace thanks to visual control. When visual and proprioceptive FB are not congruent, a conflict

appears leading to a sensory adaptation, which seems to be differently handled according to individual preferences and strategies and to the expertise level. During the learning process, visual control is used significantly at the beginning, but gradually decreases making way for a more automatic control and a change in the balance between visual and proprioceptive control. If this is the case, a possible strategy for the optimization of handwriting learning or the rehabilitation of handwriting troubles would be to facilitate the switch from control based on the written trace to control based on the movement, in order to write both precisely and fluently. The question is how supplementary FB may help with that.

SUPPLEMENTARY SENSORY FEEDBACK

Supplementary FB refers to additional sensory information provided to a writer, in complement to, or in compensation for, natural sensory FB. Therefore, supplementary FB can have two practical benefits: it can facilitate handwriting rehabilitation; it can help handwriting learning in children. We assume that providing supplementary FB to a proficient writer has little or no interest. Indeed, the *Optimal feedback control* model (for a review, see Todorov, 2004) suggests that the central nervous system sets up FB controllers that continuously convert sensory inputs into motor outputs, and that these are optimally tuned to the goals of the task by trading off energy consumption with accuracy constraints. Consequently, corrections of task-irrelevant errors are not only wasteful but they can also generate task-relevant errors (Wolpert et al., 2011).

In the usual protocols of handwriting rehabilitation, therapists correct handwriting mainly by examining the written trace. This is similar to what is done in the *Motor control* domain, when supplementary FB based on the Knowledge of the Results (KR) is supplied to the subject (Schmidt and Lee, 2005). Another possibility is to provide supplementary FB about the handwriting movement itself. This method is referred to as Knowledge of Performance (KP). Contrary to KR, which is provided after the performance of an action, KP can be provided either after or during the performance. Note that sometimes therapists give on-line FB, when they give a verbal comment about the velocity of the arm or when they hold the writer's hand to mimic the expected movement in order to make them feel the correct movement. In the present review, we will only discuss the latter option: supplying writer with real-time supplementary FB during the execution of handwriting movement in order to improve their control and aid in their rehabilitation.

Supplementary real-time FB makes it possible to change or provide additional information that a writer can access, or to supply the writer with information not naturally accessible. In other words, it can be used to amplify existing FB or give new information, not directly supplied by this FB. Although using supplementary FB to improve handwriting movements is not a recent idea (e.g., Søvik and Teulings, 1983), only recently have the increasing capacities of writing tools (e.g., graphic tablets or haptic devices) and of computers made it possible to receive real-time computer-assisted sensory FB. These new types of FB can concern the two sensory modalities already used in basic FB, vision and proprioception, and another sensory modality, audition, not naturally involved in handwriting.

SUPPLEMENTARY VISUAL FEEDBACK

Real-time visual FB has seldom been tested in handwriting because it has several limits. First, adding supplementary visual information, in a task where vision is already used to control the trace, increases the difficulty for the writer. It requires some sharing of attention that may be detrimental, especially at the beginning of learning. This was, for instance, a criticism leveled at lined paper, which adds a supplementary visual element that beginners have to cope with in addition to forming the letters they are tracing. Such a supplementary visual cue might actually compromise legibility (Weil and Amundson, 1994). Secondly, perceiving and processing visual information requires too much time to be compatible with the fast corrections that occur during handwriting (Teulings and Schomaker, 1993). Consequently, supplementary visual FB tends to slow down handwriting and to make it dysfluent, as demonstrated by Portier and van Galen (1992). Thirdly, supplementary visual cues might modify the nature of the task, transforming it into a copy task not requiring the same cognitive processes (Gonzalez et al., 2011). Finally, as already mentioned, vision is very informative concerning the spatial features of handwriting (the correctness of the letters, their position on the paper and position relative to each other. . .). However, vision is not the best sensory modality for informing optimally about the dynamic features of handwriting.

Nevertheless, if therapists do really want to add visual FB, we would advance three possibilities: First, this FB should be displayed after and not during the ongoing movement (Portier and van Galen, 1992). Supplementary visual FB on the velocity and the smoothness of the movement has been shown to be efficient when distributed after the movement (Søvik, 1981; Søvik and Teulings, 1983). The results showed that this supplementary FB improved the writing speed without diminishing accuracy, but the smoothness did not change significantly, probably because of methodological reasons. Indeed, the smoothness index was computed on the basis of the absolute velocity variability. The normal fluctuations of absolute velocity, resulting from fluctuations of curvature in the trajectory formation (Viviani and Terzuolo, 1982; Lacquaniti et al., 1983), were not taken into account.

Another possibility could be to profit from the variety of information conveyed by vision, for instance by changing in real-time the color of the ink according to a given kinematic variable (e.g., velocity or fluency). This is easy to do, from a technical point of view, thanks to graphic tablets equipped with a tactile screen. One should, however, ensure that the kinematics do not differ too much between paper and tactile screen surfaces. We would not recommend using a digitalized tablet connected to an external screen for two reasons: first, such a procedure implies that the visual FB and the proprioceptive feedback do not coincide anymore in space; secondly the change from a horizontal to a vertical plan for visual FB requires a visuomotor adaptation (grossly equivalent to a mental rotation). Both changes may affect handwriting control, especially in children with learning difficulties.

Finally, instead of adding supplementary information, another possibility could be to partially reduce visual FB. For example, with a graphic tablet it is possible to suppress the visual trace (but to preserve the vision of the pen and of the useful spatial cues like the

position on the page, the line break. . .) and thus to let the writer focus on their movement. This could be a good way to prevent the writer from paying exclusive attention to the visual trace.

SUPPLEMENTARY PROPRIOCEPTIVE FEEDBACK

Applying supplementary proprioceptive FB may be the most intuitive way of helping the writer to perceive the correct movement. This is to some extent what teachers do when they hold a child's hand and drive it along the correct trajectory in order to make him/her produce and perceive the correct movement. Applying proprioceptive FB requires the use of a mechanical device able to guide the hand of the writer: hence the generic term of *haptic guidance*. The haptic devices used in handwriting were usually multi-jointed robot arms that produce forces and allow a positioning of the pen anywhere in its workspace (see for instance Bluteau et al., 2008). A strong requirement of this method is the necessity of an *a priori* model of the ideal trajectory that the writer should reproduce. Thanks to force FB devices, haptic guidance not only informs on the current movement but also actively corrects it in relation to positional, kinematic or force errors. The question is thus to identify what the best haptic guidance is between a correction based on spatial error, focusing on the correct shape, and a correction based on force error, focusing on the correct movement.

The first studies devoted to haptic guidance were conducted on Japanese (Henmi and Yoshikawa, 1998; Yoshikawa and Henmi, 2000) and Chinese handwriting learning (Teo et al., 2002). Although they were promising, the results of these first attempts were not supported by any statistical analyses. Moreover, the learner was passively driven by the robot along the correct trajectory previously recorded by the teacher. The learner had no latitude to wander from this imposed trajectory. Therefore the FB was imposed on the passive writer. Thanks to the *Reactive Robots System* (Solis et al., 2002), the possibility appeared of taking the writer's performance into account by modifying the force FB in real-time. The Reactive Robots System not only reproduced the model, but also identified the character initiated by the writer among a panel of memorized characters and then adapted the force FB to the trajectory corresponding to the identified character. Again, the effect of this tool was not evaluated under strict experimental conditions and criteria. In particular, a comparison with a control group who did not benefit from the device was not made and therefore drawing any conclusion about the effectiveness of this method is difficult.

More recently, haptic guidance was evaluated under more rigorous experimental conditions for handwriting learning (Palluel-Germain et al., 2007). The authors evaluated the effect of a visuo-haptic device in 5 to 6 year-old children carrying out a copying task. Kinesthetic FB was based on positional error, by comparing the produced trajectory and the nearest point of the model trajectory. They found that children who learned with the visuo-haptic device exhibited more fluent movements. Interestingly, they concluded that augmented kinesthetic FB in such a learning protocol increased the proactive strategy of the child's control. One year later, Bluteau et al. (2008) tested the efficiency of two kinds of haptic guidance in adults, based on position or force errors. The position error corresponded to the Euclidean

distance between the ideal trajectory required by the task and the produced trajectory. The force error corresponded to the difference between the force produced by the user and the force used in the model guiding the robot for the theoretical trajectory. These last authors got a positive effect of haptic guidance based on force error alone.

In conclusion, applying haptic guidance for changing proprioceptive FB seems relatively promising, provided that the guidance is based on a dynamic error and not on spatial error. However, its effectiveness remains to be confirmed because this device has been evaluated mostly on simple motor tasks (for a review, see Sigrist et al., 2013), but to a lesser extent on more complex tasks like handwriting. In addition, haptic guidance necessitates specific devices that can be costly and complex to use. Furthermore, as already explained, proprioceptive guidance with force FB devices requires an initial recording of an 'ideal' trajectory that the writer would then have to reproduce and from which corrections could be made. The individuality and variability of poor handwriting brings into question the validity of methods based on dynamic model reproduction for rehabilitation. This question has not been addressed until now.

SUPPLEMENTARY AUDITORY FEEDBACK

Sounds can be used to add supplementary information that the writer does not take into account or that he/she cannot access naturally (e.g., inform about the muscular activity, Ince et al., 1986). It can also be used as an alternative channel of information processing in order to compensate for a deficit in another sensory modality (e.g., compensate for a visual deficit by giving spatial information, Plimmer et al., 2011). Until now, no study has investigated the role of audition in handwriting motor control. Handwriting has always been considered as a silent activity. Because there is no link *a priori* between handwriting and sounds, auditory FB could be used to inform on many different variables. The question is thus to discover how and on what, sounds may help in improving motor control or the relearning of handwriting.

Historically, auditory FB in handwriting has only been used to treat one particular neurological deficit: namely writer's cramp. Reavley (1975) was the first who tried to transform EMG activity into sounds in order to supply patients with auditory biofeedback. He wanted to help them to better contract muscles that were not appropriately activated. The author reported improvements in terms of "quick, effective and legible handwriting". However, he did not describe the apparatus and auditory FB used. Other attempts to inform patients about their muscular activity were made with auditory FB varying in intensity as a function of EMG activity. Bindman and Tibbetts (1977) treated six patients over periods ranging from 3 months to 5 years. Auditory FB consisted of increasing or decreasing sound intensity, depending upon muscle contraction or relaxation. The authors reported that one patient became completely symptom-free, one improved sufficiently to produce little or no disability at work, two improved but continued to have some work disability and two evidenced no change. However, as reported by Ince et al. (1986), no details were provided in this article on any aspect of methodology, apparatus, muscle activity, or data analysis. No pre- or post-treatment handwriting samples were included for visual inspection of the changes. In

another study (Cottraux et al., 1983) the auditory biofeedback consisted of an analog audio signal that the patients had to reduce in pitch by decreasing the tension of their muscles. Again, no statistical analyses were performed and several methodological criticisms can be raised (Ince et al., 1986 for more details). O'Neill et al. (1996) reported a case of a patient whose symptoms disappeared after one week of such treatment. Such rapid effects were surprising and Deepak and Behari (1999) made another attempt on ten patients with Writer's Cramp and hand dystonia. They revealed that nine of them showed an improvement of between 37 and 93% in handwriting after daily practice with auditory biofeedback over a few months. Nevertheless, although the technique seemed relevant, the biofeedback from EMG activity was questioned by Ince et al. (1986) and by Deepak and Behari (1999) who admitted that the first results were not totally convincing. One of the limitations was that handwriting involves many muscles, sometimes small and profoundly located, whose activity is difficult to record. Possibly, more invasive methods (e.g., intra-muscular EMG) would overcome this problem, but such methods remain difficult to use and restricted to serious neuromuscular pathologies.

Muscular activity is directly linked to forces exerted by the muscles, and patients suffering from writer's cramp are known to apply too great a force with their fingers onto the pen. Therefore, transforming the grip force into sounds has been tested as a way of leading patients to decrease the force of their grip (Baur et al., 2009). The pen was equipped with force sensors and the auditory FB consisted of a continuous low-frequency tone when the average grip force exceeded 5 N. The tone frequency increased in four steps with the grip force level and patients were instructed to perform the writing exercises in such a way that they heard a pleasant, low-frequency tone. After several hours of training, the authors reported that both the grip force and the vertical pressure applied by the pen on the paper decreased in the patients. This easy to apply and non-invasive method seems quite encouraging for the rehabilitation of writer's cramp.

In addition to a simple association between a physiological variable and a sound, such as those previously described, more complex associations can also be used when the goal is to supply auditory FB about movements: This is the so-called movement sonification (Effenberg, 2005; see Sigrist et al., 2013 for a review). In the case of handwriting, the purpose is to enrich perception by adding auditory signals linked to given variables of handwriting movement (spatial, kinematic, or dynamic information). According to the values of the chosen variables, one or several sound parameters can be modified. As Sigrist et al. (2013) observed, however, it is fundamental to define as precisely as possible the sonification strategy. Two issues have to be solved: the '*what to sonify?*' consists of identifying precisely which variables should be sonified, i.e., the sound mapping; the '*how to sonify?*' consists of evaluating the auditory design. Now, with regards to handwriting, there is a lack of solutions to these problems in the current literature.

Is auditory FB relevant for informing about the spatial characteristics of handwriting? Andersen and Zhai (2010) made a first attempt at answering this question. They compared both the accuracy of the written trace and the speed of execution under four

conditions resulting from the crossing of two types of FB: with and without visual FB on the written trace, and with and without supplementary auditory FB linked to the pen's position. They related a positive effect from supplementary auditory FB on the motivation of the learners, but no direct influence on their performance. This absence of effect can be explained by the fact that spatial information is harder to translate into the auditory than into the visual dimension (Welch, 1999).

On the other hand, the dynamic features of sounds make them particularly appropriate for signaling the movement's dynamics. Indeed, when listening carefully to the noise produced by handwriting, one can hear a friction sound generated by the pen–paper interaction, especially when the surface is rough. This friction between the pen tip and the paper's asperities produces sound variations related to the handwriting kinematics that may, to a certain extent, inform on what the writer is writing. Thoret et al. (2014) tested this hypothesis with a synthetic friction sound whose timbre variation was related to the pen's velocity. In a first task, they demonstrated that the timbre variations produced from the sound of a moving pen appear to vary in accordance with the kinematic rule governing real graphical movements. In a second experiment, the authors investigated the ability to categorize drawn shapes 'by ear'. Subjects were asked to associate friction sounds with simple graphic shapes. They concluded that categorization of visual shapes on the basis of their produced sounds was possible if the kinematics differ sufficiently. However, these results were acquired using simple graphic shapes that had been drawn by the very fluid movements of an adult writer. Contrary to drawing simple shapes, handwriting imposes complex movements which are not always fluid, in particular for handwriting learning or rehabilitation.

(Danna et al., 2013a,b, 2014) have studied the effect of movement sonification for handwriting learning and rehabilitation. The sonification strategy consisted of using an intuitive mapping between sound and movement, i.e., friction sounds that might have been naturally created by real pen movements. The timbre of the sounds varied with the instantaneous velocity of the pen (see Thoret et al., 2014). To evaluate the potential of the technique, these authors carried out a series of experiments. The first experiment was designed to answer the question: "Is it possible to identify poor handwriting only by ear, without seeing the written trace?" (Danna et al., 2013a). In a pre-experiment, samples of the same word written on a graphic tablet by children with dysgraphia, children with proficient handwriting, and proficient adult writers were collected. Then, from these samples, three handwriting variables – the instantaneous velocity, the movement fluency, and the axial pen pressure – were sonified in order to create audio files which were then played to naïve adult listeners who had to mark the quality of the underlying unseen handwriting. The listeners were not aware that the sounds corresponded to three different groups of writers. The results showed that, when they were informed about the meaning of the sounds and the evaluation criteria, all listeners marked the dysgraphic handwriting lower than that of the two other groups. So it appeared possible to discriminate only by ear between proficient and poor handwriting. This result validated the sounds used for informing on the quality of handwriting. However, the sounds here were not FB: they were

not played back to the person whose handwriting movements had generated them.

The same sonification strategy was applied as real-time auditory FB for improving handwriting learning (Danna et al., 2014) and rehabilitation (Danna et al., 2013b). Auditory FB improved the learning of new characters in adults with their non-dominant hand in a single training session (Danna et al., 2014). A positive effect was also obtained in a rehabilitation protocol lasting several weeks involving children with dysgraphia (Danna et al., 2013b). However, the performance increase with sonification should be compared to a control situation where children do not benefit of the sonification, and this was not done in this pilot study.

In conclusion, applying concurrent auditory FB seems very promising, provided that the auditory FB informs about handwriting movement and not about a spatial characteristics. Indeed, vision is more appropriate than audition for perceiving spatial information, whereas sounds can naturally reveal phenomena containing dynamic cues to which the eye is less sensitive (e.g., Fitch and Kramer, 1994). Moreover, auditory FB may be efficient without leading the learner to become dependent on external FB. Ronsse et al. (2011) demonstrated that learners are less dependent on auditory than on visually augmented FB. Because audition is available during handwriting, sounds may be used to complement visual and proprioceptive FB and enlighten the writer about “hidden” dynamic variables which are not sufficiently taken into account, particularly at the beginning of learning or in rehabilitation. However, the efficiency of auditory FB depends considerably on its correct interpretation, since listening to auditory displays is less common than viewing visual displays (Sigrist et al., 2013). Finally, in addition to their informative characteristics, sounds can be fun and can motivate learners. Since handwriting learning or rehabilitation requires daily training over several months, the learner’s motivation is one of the most important components to take into account.

Another possibility consists of applying multimodal concurrent FB. To our knowledge, only one study has proposed a multimodal system based on coupled auditory and haptic FB to help blind children to sign (Plimmer et al., 2011). They showed that with such multisensory FB, blind children more quickly learned to sign. No precise information regarding the kinematic variables was reported in this clinical study.

CONCLUSION

The mastering of handwriting is so essential in our society that it is important to try to find new methods for facilitating its learning and rehabilitation. With the technical means we now have at our disposal, supplying writers with new types of sensory FB that are richer than those naturally used, is easily conceivable. We have just recalled that two sensory modalities are involved in handwriting control: vision and proprioception, and they do not inform on the same aspects of handwriting. Vision is more suited for checking the quality of the written trace and proprioception for controlling the ongoing movement. Providing enriched FB in each of them is theoretically possible, however, these types of FB should respect their specificities. In particular, a new visual type of FB can hardly be provided during the execution of the movement without the risk of overloading the cognitive capacities and inducing

a subsequent degradation of the movement. Proprioceptive supplementary FB is likely to be more appropriate to facilitate the execution of a fluent movement, however it can be costly and is not easy to use. Finally, another sensory modality, namely audition, which does not naturally contribute to handwriting control, could be a good candidate for adding supplementary FB. Audition has several advantages: first, supplementary auditory FB is less likely to overload the cognitive process than additional visual FB, second it is particularly suited to informing about the unfolding of a process and hence about the movement kinematics, third it is easy to use, and finally it might add an element of play to the learning process, particularly for children. Enriched handwriting based on new multisensory FB is also conceivable. Using real-time supplementary FB to assist in the handwriting process is probably destined for a brilliant future with the growing availability and rapid development of tablets.

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The differential time course for consonant and vowel processing in Arabic: implications for language learning and rehabilitation

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Educators and therapists in the Arab world have not been able to benefit from the recent integration of basic behavioral science with neuroscience. This is due to the paucity of basic research on Arabic. The present study is a step toward establishing the necessary structure for the emergence of neuro-rehabilitory and educational practices. It focuses on the recent claim that consonants and vowels have distinct representations, carry different kinds of information, and engage different processing mechanisms. This proposal has received support from various research fields, however it surprisingly stops short of making any claims about the time course of consonant and vowel processing in speech. This study specifically asks if consonants and vowels are processed differentially over time, and whether these time courses vary depending on the kind of information they are associated with. It does so in the context of a Semitic language, Arabic, where consonants typically convey semantic meaning in the form of *tri-consonantal roots*, and vowels carry phonological and morpho-syntactic information in the form of *word patterns*. Two cross-modal priming experiments evaluated priming by fragments of consonants that belong to the root, and fragments of vowels belonging to the word pattern. Consonant fragments were effective primes while vowel fragments were not. This demonstrates the existence of a differential processing time course for consonants and vowels in the auditory domain, reflecting in part the different linguistic functions they are associated with, and argues for the importance of assigning distinct representational and processing properties to these elements. At broader theoretical and practical levels, the present results provide a significant building block for the emergence of neuro-rehabilitory and neuro-educational traditions for Arabic.

Keywords: consonantal root, vocalic word pattern, time course of spoken word processing, CV-hypothesis, learning and rehabilitation

INTRODUCTION

Neuro-rehabilitation and neuro-education are two nascent scientific disciplines that are informed by research from cognitive neuroscience and behavioral psychology (Taub et al., 2002; Devonshire and Dommett, 2010; Ansari et al., 2012; Hook and Farah, 2012). The main aim of neuro-rehabilitation is to ameliorate dysfunctional cognitive and brain functions caused by disease or injury (Robertson and Fitzpatrick, 2008; Nehra et al., 2014). In contrast, neuro-education seeks to create a better understanding of how we learn and how knowledge about the functional properties of the brain can be harnessed to create more effective teaching methods, curricula, and educational policies (Hardiman et al., 2009; Carew and Magsamen, 2010). Despite their relatively recent history both disciplines are making significant strides toward helping with rehabilitory and educational processes. This success has been made possible thanks to the burgeoning fields of cognitive neuroscience and behavioral psychology. For example, recent research has revealed the existence of

“neural markers” of learning disorders, most notably in the case of dyslexia. Imaging studies have revealed that human infants at risk of dyslexia (i.e., with immediate family members who suffer from dyslexia) show atypical neural responses to changes in speech sounds, even before they are able to understand the semantic content of language (Leppänen et al., 2002). Such a finding allows for the early identification and remediation of potential learning disorders.

Unfortunately however the blossoming of cognitive neuroscience and behavioral psychology holds true only of certain regions in the world such as Europe and North-America. Other geographic areas, in particular the Arab world, suffer from a pronounced dearth of basic research which has led to serious deficiencies in effective interventions in neurorehabilitation and a complete lack of a neuro-educational culture.

The purpose of the present paper is to make an initial contribution to developing the necessary building blocks from basic psycholinguistic research for the emergence of neuro-rehabilitory

and neuro-educational practices in the Arab world. More specifically, the paper reports two psycholinguistic experiments aiming at determining how information about consonants and vowels is derived from the auditory input and mapped onto lexical knowledge. The results, as we will argue in the general discussion, can inform practitioners both in rehabilitation and education.

CONSONANTS AND VOWELS IN LANGUAGE PROCESSING

A long-standing debate in cognitive science relates to what levels of representations are available to and used by the language processing system. In the context of this general debate, many studies have come to focus on the status of consonants and vowels, asking whether these are categorically distinct objects that are independently represented and differently processed (Caramazza et al., 2000; Nespor et al., 2003; Bonatti et al., 2005, 2007; Nazzi, 2005; Mehler et al., 2006; Knobel and Caramazza, 2007; Toro et al., 2008), or whether they are simply convenient labels that distinguish sonority peaks (vowels) from sonority troughs (consonants) in the speech stream and need neither to be interpreted as distinct constructs, nor to invoke different processing mechanisms (Monaghan and Shillcock, 2003, 2007; Keidel et al., 2007).

The existing data relating to this debate derive exclusively from research into Indo-European languages. Caramazza et al. (2000) report data from two Italian patients who showed contrasting selective difficulties in producing vowels and consonants. These results were taken as evidence that consonants and vowels are independently represented. However, this interpretation was challenged by Monaghan and Shillcock (2003, 2007), who argued that the double dissociation between vowels and consonants can be modeled as an emergent effect of modular processors operating on feature-based representations with no need to posit separate representations for vowels and consonants.

Bonatti et al. (2005) report a study in which French speaking subjects learned an artificial language where words were strings of alternating consonants (C) and vowels (V) (e.g., *puragi), and were asked to indicate in a forced choice test which items belonged to the artificial language. Subjects picked up on the regularities for consonants, but not vowels suggesting that the two elements engaged different processing mechanisms. Keidel et al. (2007) challenged this interpretation and contended that differences in the distribution of consonants and vowels in French may explain why French speakers pick up on the regularities provided by consonants but not those provided by vowels.

Keidel et al.'s criticism was addressed in a subsequent study by Toro et al. (2008), who extended Bonatti et al.'s findings to Italian speakers. Specifically, having learnt a nonsense set like *badeka, *bedake, where the vowel structure is ABA, Italian speakers preferred sequences like *biduki, *budiku, with the same ABA vowel structure, although the vowel sequences -i-u-i or -u-i-u were never part of the familiarization stream. Importantly, the same subjects were unable to extract comparable generalizations using consonants from sequences like *benobu and *pikeko.

The distinction between consonants and vowels at the cognitive level has some support at the neural level. For instance,

neuropsychological research offers descriptions of lesions in left temporal, parietal and fronto-parietal regions or bilateral parietal cortex which affect consonants and vowels differentially (Caramazza et al., 2000). Similarly, electrophysiological evidence broadly suggests anterior-posterior dissociation for consonants and vowels, respectively. For instance, Carreiras et al. (2007) showed that correct NO responses to pseudowords during lexical decision evoked N400 effects in anterior (F5 line) and middle regions (C5 line) when consonants are transposed, but in middle and posterior regions (P5 line) when vowels are transposed. However, there was no clear lateralization associated with these effects. A more fine-grained localisation is provided using PET (Sharp et al., 2005). In Sharp et al.'s study participants were asked to generate real words from heard pseudowords created by the substitution of either a vowel or consonant. When consonants needed to be substituted, word generation was more difficult and left inferior frontal activation was higher. However, there was no increase of activation for vowels relative to consonants in the left suggesting that that vowel processing may share neural resources with prosodic processing in the right hemisphere. More recently Carreiras and Price (2008) used functional magnetic resonance imaging to investigate whether vowel and consonant processing differences are expressed in the neuronal activation pattern and whether they are modulated by task. The tasks used were reading aloud and lexical decision on visually presented pseudowords created by transposing or substituting consonants and vowels in real words. In the reading aloud task, changing vowels relative to consonants increased activation in a right middle temporal area typically associated with prosodic processing of speech input. In contrast, in the lexical decision task, changing consonants relative to vowels increased activation in right middle frontal areas typically associated with response inhibition. The task-sensitive nature of these effects underscores the fact that consonants and vowels place differential processing demands on the brain and differentially engage various neural structures.

These results provided the basis for the development of the Consonant Vowel-hypothesis (CV-hypothesis) which holds that consonants and vowels fulfill different roles across languages with consonants carrying lexical information and vowels encoding morpho-syntactic and phonological information. These two elements also engage two processing mechanisms. The first relies on transitional probabilities between consonants to extract words and access the lexicon for meaning, while the second relies on the structure defined by vowels to draw generalizations about the input (Nespor and Vogel, 1986; Nespor et al., 2003; Bonatti et al., 2005; Nazzi, 2005; Toro et al., 2008).

The CV-hypothesis has far-reaching implications. Not least, it implies that the language system consists of different levels of representations each with its own internal structure, and each with its specialized computational requirements. Although these implications impose stringent constraints on the dynamics of online spoken word recognition, the CV-hypothesis remains highly underspecified in this respect. Specifically, it says nothing about the potential timing differences underlying the uptake of information about consonants and vowels from speech. For the proper development of the CV-hypothesis as a speech processing

model, it is essential to specify the dynamics of the speech mapping process.

The present paper aims to fill this gap (a) by developing, within the CV-hypothesis framework, a set of specific claims about the time course underlying the projection of consonantal and vocalic information onto lexical representations, and (b) by empirically testing these claims. To do this, one needs to evaluate consonants and vowels in languages where the distinction between these two elements is not limited to the phonological domain, but has overt implications for meaning. Semitic languages like Arabic and Hebrew offer this opportunity, with the consonant-vowel contrast being overtly relevant at morphological, semantic and syntactic levels (McCarthy, 1981). Consequently this study focuses on Arabic root consonants as bearers of semantic meaning, and vocalic patterns as carriers of phonological and morpho-syntactic information. In so doing it will take our understanding of the cognitive architecture subserving consonant-vowel processing to a new level of generality by exploring the Semitic system, while achieving at the same time new levels of specificity by uncovering potentially different processing procedures across languages.

CONSONANTS AND VOWELS IN A SEMITIC CONTEXT

The consonant-vowel contrast in Semitic languages defines two functionally distinct morphemes, with consonants making up the *root*, and vowels corresponding to the *word pattern*¹. The root is typically comprised of 3 consonants and conveys the general meaning which will be present to various degrees in all other words featuring that root. By contrast the word pattern is a composite morpheme with derivational and inflectional functions (Prunet et al., 2000; Boudelaa and Marslen-Wilson, 2004, 2005). More specifically, the word pattern is like a template that determines not only the overall shape of the surface form, its phonological structure and its stress pattern, but it is also the bearer of morpho-syntactic information such as *active*, *passive*, *plural*, etc. Both the root and the word pattern are bound morphemes and cannot surface unless they are interleaved within each other nonlinearly. For example the Arabic root {ktm} with the general meaning of *hiding*, and the pattern {-a-a-} with a *past tense active* meaning are interleaved to generate the surface form [katam] *hide*, *active*, *perfective*, *verb*.

In such a linguistic environment, it makes sense that consonants and vowels are segregated since downstream processing relies on such distinct parts of the word. This raises the question of whether this segregation results in a differential uptake of information about roots (consonants) and word patterns (vowels) over time.

THE CV-HYPOTHESIS IN A SEMITIC CONTEXT

Spoken word recognition is a rapid process, typically taking less than a quarter of a second to complete (Pulvermüller et al., 2006; Hauk et al., 2009). For this reason, it is generally thought that the process of mapping speech input onto internal representations is governed by the principle of maximal processing efficiency,

whereby the incoming speech is analyzed at all points and the most informative output available is derived from it (Marslen-Wilson and Welsh, 1978; Marslen-Wilson and Tyler, 1980; Pirog Revill et al., 2008a,b). If, as the CV-hypothesis claims, the recognition process is oriented toward consonants which are used to extract information about word identity, consonants should be processed continuously such that at each point in time where the speech input contains a consonant, a lexical access process is initiated and the best fitting candidates are activated. In the context of priming, this means that a fragment of an Arabic root should be an effective prime of a target sharing the same root consonants. This prediction is tested in Experiment 1.

Another important feature of the CV-hypothesis is that vowels are used to derive generalization about the linguistic environment. Generalization is the notion that humans are able to respond in a similar way to different stimuli provided those stimuli have similar properties. In this respect, evoking the same response to the Arabic words [katab] *write*, and [daxal] *enter*, based on their similar vowel patterns is an instance of generalization. Arguably, in order to derive this kind of generalization one needs to hear the full sequence. If correct, this predicts that information about vowels should not be mapped continuously because the language processor needs to accumulate enough information before it can draw reliable generalizations about the overall structure of the word (Lahiri and Marslen-Wilson, 1992). In a priming context, this translates into the prediction that a fragment of an Arabic word pattern should be less effective as a prime than the full word pattern. Experiment 2 tests this prediction.

EXPERIMENT 1

Is information about root consonants mapped continuously onto internal representations of lexical form as the CV-hypothesis predicts? This question is tackled using the cross-modal priming paradigm in which participants have to make a speeded lexical decision about a visual target presented immediately at the offset of an auditory word-fragment prime or a full word prime. Since the prime is auditory and the target is visual in this paradigm, any savings in the processing of the target should be attributed to repeated access of the same underlying modality-independent representation. If priming obtains between words sharing a root fragment, this will suggest that the consonantal root is represented as an independent unit, and that information about it is continuously evaluated, segment by segment as that information becomes available.

METHOD

Participants

Eighty one volunteers (50 females) aged 16 to 20 were tested. They were students at the high school of Tataouine in the South of Tunisia. The subjects were native MSA speakers and studied French and English as second and third languages. None of them had any history of hearing loss or speech disorders. Written consent to take part in the study was obtained either from the participants themselves or from their guardians if they were minors. The study was approved by the Peterborough and Fenland Ethical Committee.

¹ Although the root is exclusively consonantal, the word pattern may feature a subset of consonants along with its vowels.

Materials and design

Forty-eight orthographically unambiguous targets were used. They were on average 4.58 letters long (*SD*: 0.66), 8 phonemes long (*SD*: 1.25), and 3.33 syllables long (*SD*: 0.48). Ten different word patterns were used to construct this set of words, which was divided into two subsets of 24 words each, matched on length and frequency checked using the ARALEX database (Boudelaa and Marslen-Wilson, 2010). Each target in the first set was paired with four types of primes as outlined in **Table 1**.

In the +Root, Full Prime condition, the priming word (e.g., [buluuxun] *puberty*), and the target (e.g., [baliixun] *eloquent*) share the consonants of the root {blx} and the prime is presented in full. In condition 1b, labeled +Root, Partial Prime, the same target [baliixun] is paired with the fragment [buluu] excised from the full prime [buluuxun]. Note that the only shared material across prime and target are consonants. To provide appropriate controls against which to measure priming, the target [baliixun] was paired with an unrelated full prime [tʰumuuhun] *ambition*

Table 1 | Examples of stimuli used in the different conditions of experiment 1.

	Prime	Target
1a: +Root, Full Prime	بلوغ [buluuxun] <i>puberty</i>	بليغ [baliixun] <i>eloquent</i>
1b: +Root, Partial Prime	بلو [buluuxun]	بليغ [baliixun] <i>Eloquent</i>
1c: Baseline, Full Prime	طموح [tʰumuuhun] <i>Ambition</i>	بليغ [baliixun] <i>Eloquent</i>
1d: Baseline, Partial Prime	طمو [tʰumuu]	بليغ [baliixun] <i>eloquent</i>
2a: +Phon, Full Prime	بلوغ [buluuxun] <i>puberty/reaching</i>	بليد [baliidun] <i>silly</i>
2b: +Phon, Partial Prime	بلو [buluu]	بليد [baliidun] <i>silly</i>
2c: Baseline, Full Prime	طموح [tʰumuuhun] <i>Ambition</i>	بليد [baliidun] <i>silly</i>
2c: Baseline, Partial Prime	طمو [tʰumuu]	بليد [baliidun] <i>silly</i>

Examples are given in Arabic script with a phonetic transcription and an English gloss.

in the Baseline Full Prime condition and with the unrelated fragment [tʰumuu] taken from the full prime [tʰumuuhun] in the Baseline Partial Prime condition.

To ensure that the partial primes are ambiguous and can be the beginning of different words in the language, the first three to four segments of each full prime in conditions 1a and 2a were excised and presented in a simplified gating task to 15 subjects from the same linguistic background and same age range as those who took part in the priming experiment (Grosjean, 1980). In this task subjects are typically presented with successive auditory fragments of 50 ms long, and they are instructed to suggest a word that can be a continuation to the fragment being presented, and to say how confident they are of their guess. Each partial prime was on average 175 ms long (*SD*: 25.31), and was presented incrementally in steps of 50 ms. At each step subjects had to guess the word and to say how confident they were of their guess on a scale from 10 *completely sure* to 1 *completely unsure*. For each fragment, subjects suggested on average 11.41 possible different words, (8.12 different roots), and their confidence ratings (their degree of certainty about the word they suggested) were low, averaging 4 on a 10-point scale. Taken together the large number of suggested words and the low confidence ratings suggest that the fragment of the word the subjects were exposed to was ambiguous enough to match or activate different possible lexical candidates.

The 24 targets in the second set were also paired with the same set of primes to form a phonological control condition. In other words, the primes were constant but the targets were different. Phonological overlap is defined in this study as the number of shared phonemes from onset between a given pair of words. This is illustrated in the +Phon, Full Prime and the +Phon, Partial Prime conditions where the word [buluuxun] *puberty*, and its fragment [buluu] are used to prime the phonologically related target [baliidun] *silly*. This target is phonologically related to the partial prime [buluu] and provides a viable continuation to its consonantal structure. However, it features the root {bld}, which is different from the root {blx} underlying the full-prime [buluuxun] *puberty*. The control prime [tʰumuuhun], used in the Baseline, Full Prime condition, and its fragment [tʰumuu] used in the Baseline, Partial Prime condition, provide baseline conditions for evaluating priming in the +Phon, Full Prime and the +Phon, Partial Prime.

Primes and targets shared on average 57.3% of their phonemes in condition 1a condition, 44.6% in condition 1b, 56.3% in condition 2a, and 47.9% in condition 2b. Seventy-two unrelated word-word pairs were included to reduce the proportion of related pairs in the experiment to 20%. Half of these had a partial prime and half a full prime. Another 120 word-nonword pairs with similar characteristics as the word-word pairs were used to provide the nonword targets needed for the lexical decision task employed here. Forty practice trials that were representative of the experimental trials were used. To avoid repetition of primes and targets within subjects, four counterbalanced experimental lists were constructed each consisting of 280 pairs.

Procedure

The prime words were recorded by a native speaker of Arabic and digitized with a sampling rate of 44 kHz. Subjects heard

the stimuli at a comfortable level through HD 250 Sennheiser headphones. The sequence of stimulus events within each trial started with a 1000 ms silence followed by an auditory prime. Immediately at the offset of the prime a visual target was displayed on the screen for 2000 ms. Timing and response collection were controlled by a laptop PC running the DMDX package (Forster and Forster, 2003). Participants were instructed to make a lexical decision as quickly and as accurately as possible. The experiment, which lasted for 35 min, started with the practice trials followed by the rest of the stimuli.

RESULTS AND DISCUSSION

Error trials were excluded and not replaced (2.66%). Outlying responses above 3000 ms or below 100 ms were also excluded (0.07%). The remaining data were inverse transformed (multiplied by 1/1000) to reduce the influence of outliers (Ratcliff, 1993). **Table 2** gives the percent error rates and the means of the reaction times.

Two mixed design analyses of variance (ANOVAs) across subjects (F1) and across items (F2) were conducted on the reaction time and accuracy data. They included the factors Condition (morphology vs. phonology), Prime Type (related vs. unrelated), and Prime Length (full prime vs. partial prime). Condition was treated as a repeated factor in the participants' analysis and as an unreplicated factor in the items analysis, while Prime Type was treated as a repeated factor in both analyses. A fourth variable "List" was also included in the analyses as a dummy variable to reduce the estimate of random variation (Pollatsek and Well, 1995). This variable was treated as a between-subjects factor in the participants analysis and as a between-items factor in the items analysis. The *p*-values reported for the ANOVA in this and the next experiment are adjusted with the Greenhouse–Geisser epsilon correction for nonsphericity. There were significant main effects of the factors Condition [$F_{1(1,80)} = 37.43, p < 0.0001$; $F_{2(1,47)} = 10.52, p < 0.0022$] and Prime Type [$F_{1(1,80)} = 59.16, p < 0.001$; $F_{2(1,47)} = 14.76, p < 0.001$]. The main effect of Prime Length was not significant [F_1 and $F_2 < 1$]. Condition interacted significantly with Prime Type [$F_{1(1,80)} = 25.14, p < 0.001$; $F_{2(1,47)} = 17.71, p < 0.05$], and Prime Length [$F_{1(1,80)} = 18.38, p < 0.001$; $F_{2(1,47)} = 15.76, p < 0.005$]. The two-way interaction between Prime Type and Prime Length was not significant [$F_1 < 1$; $F_2 < 1$]. The theoretically important three-way interaction between Condition, Prime Type and Prime Length was significant [$F_{1(2,80)} = 6.18, p < 0.01$; $F_{2(2,47)} = 10.52, p < 0.01$], indicating that priming was not constant across the different

conditions. Further planned comparisons using 0.05 Bonferroni protection levels confirmed this (Keppel, 1982). Priming was significant in the [+Root, Full Prime] case [$F_{1(1,80)} = 49.36, p < 0.001$; $F_{2(1,23)} = 10.94, p < 0.001$], the [+Root, Partial Prime] case [$F_{1(1,80)} = 39.83, p < 0.001$; $F_{2(1,23)} = 8.61.52, p < 0.001$], and the [+Phon, Partial Prime] case [$F_{1(1,80)} = 7.27, p < 0.05$; $F_{2(1,23)} = 6.65, p < 0.05$], but not in the [+Phon, Full Prime] case [$F_1 < 0.05$; $F_2 < 1$]. Furthermore, the magnitude of priming in the [+Root, Full Prime] was not significantly different either from that in the [+Root, Partial Prime] case or the [+Phon, Partial Prime] case [all $F_s < 1$]. By contrast there was a reliable difference between the amount of priming observed in the [+Phon, Full Prime] condition and (a) the [+Root, Full Prime] condition [$F_{1(1,80)} = 6.86, p < 0.05$; $F_{2(1,23)} = 5.04, p < 0.05$], (b) the [+Root, Partial Prime] condition [$F_{1(1,80)} = 5.91, p < 0.05$; $F_{2(1,23)} = 0.059$], and (c) the [+Phon, Partial Prime] condition [$F_{1(1,80)} = 7.12, p < 0.05$; $F_{2(1,23)} = 6.18, p < 0.05$]. Similar statistical analyses were conducted on the error data but no across conditions differences were found.

Finally, to check on the possible contribution of two stimulus properties—the acoustic duration of the prime, and the degree of phonological overlap between prime and target—each of these variables was centered and used as a predictor of priming in separate stepwise multiple regression analyses. Neither duration [$R^2 = 0.003, F_{(1,94)} = 0.26, p = 0.60$] nor phonemic overlap [$R^2 = 0.004, F_{(1,94)} = 0.34, p = 0.55$] was a significant predictor of priming.

This experiment suggests that full word primes and targets sharing a consonantal root prime each other reliably, while phonologically related full primes fail to do so. This is consistent with a continuous view of spoken word recognition, but it does not rule out the possibility of discontinuous processing whereby the language processor waits until the whole three consonants of the root are heard before it attempts lexical access. This interpretation is ruled out however by the effects for partial primes. In particular, partial primes such as [buluu] are as effective as the complete word [buluuvun] in priming lexical decision to a probe with which they share the same consonants (e.g., [baliivun] *eloquent*; [baliidun] *silly*). This suggests that the lexical processor attempts to find a lexical match as soon as sufficient consonantal information is extracted from the speech stream. Upon hearing the partial prime [buluu] many word candidates whose roots contain the consonants {b,l} (e.g., {blx}, {bll}, {blɿ}, {bly}) are activated and start competing for recognition. Otherwise there would be no basis for the comparable facilitation observed in the +Root Partial Prime condition and the +Phon Partial Prime condition. In summary, the results of Experiment 1 suggest that information about the consonantal root is continuously evaluated as the relevant information becomes available in the speech stream.

EXPERIMENT 2

This experiment asks whether word patterns, like roots, are continuously mapped onto the lexicon. It co-varies two factors: (a) the prime and target relationship such that they share either a word pattern or a phonological overlap, and (b) prime length using full words or fragments of words as primes. To derive

Table 2 | Reaction times, (standard deviations), amount of priming, and %error rates for the target preceded by test and baseline primes in the different conditions of experiment 1.

Condition	Test	Baseline	Priming	Error in Test	Error in Baseline
+Root, Full Prime	557 (41)	595 (52)	38	1.74	2.23
+Root, Partial Prime	568 (47)	602 (50)	34	3.10	2.71
+Phon, Full Prime	609 (55)	604 (64)	−5	3.10	2.72
+Phon, Partial Prime	583 (53)	600 (63)	17	3.29	2.33

predictions for this experiment, two claims need to be allied. The first is the CV-hypothesis claim that vowels (word patterns) are used to draw generalizations about the structure of the input. The second is the claim that generalizations can be successfully drawn only if the appropriate information is available. Thus, the question is whether partial information about the Arabic word pattern is enough to identify the correct pattern and trigger lexical access.

METHOD

Participants

Eighty-eight volunteers (40 females) from the same age range and background as those in Experiment 1 were tested.

Materials and design

Forty eight orthographically unambiguous words were chosen to serve as targets. They consisted on average of 3.54 letters (*SD*: 0.85), 3.38 syllables (*SD*: 0.73) and 6.06 phonemes (*SD*: 1.59). Fourteen different word patterns were used to construct these items, which were divided into two sets of 24 words matched on length, and frequency, checked using the ARALEX database. Each target in the first set was paired with four types of primes as illustrated in **Table 3**.

Table 3 | Sample stimuli used in the different conditions of experiment 2.

	Prime	Target
1a: +WP, Full Prime	وقوع [wuquuʕun] <i>happening</i>	دخول [duxuulun] <i>entering</i>
1b: +WP, Partial Prime	وقو [wuquu]	دخول [duxuulun] <i>entering</i>
1c: Baseline, Full Prime	دقيق [sufunun] <i>accurate</i>	دخول [duxuulun] <i>entering</i>
1d: Baseline, Partial Prime	دقيـ [daqii]	دخول [duxuulun] <i>entering</i>
2a: +Phon, Full Prime	وقوع [wuquuʕun] <i>happening</i>	تبادل [tubuudila] <i>to be exchanged</i>
2b: +Phon, Partial Prime	وقو [wuquu]	تبادل [tubuudila] <i>to be exchanged</i>
2c: Baseline, Full Prime	دقيق [daqiiqun] <i>accurate</i>	تبادل [tubuudila] <i>to be exchanged</i>
2c: Baseline, Partial Prime	دقيـ [daqii]	تبادل [tubuudila] <i>to be exchanged</i>

Examples are given in Arabic script with a phonetic transcription and an English gloss.

In the +WP, Full Prime condition, the prime is a full word (e.g., [wuquuʕun] *happening*) that shares the vowels of the word pattern (e.g., {u-uu-} *perfective, active*) with the target (e.g., [duxuulun] *entering*). In the +WP, Partial Prime condition, the same target is paired with a fragment (e.g., [wuquu] excised from the full prime [wuquuʕun]). The fragment primes were on average 238 ms long (*SD*: 38). In a gating task run on these fragments (see Experiment 1 for details), 15 subjects who did not participate in the priming experiment suggested on average 11.97 possible different words (with 3.6 different word patterns on average). Their confidence ratings were generally low averaging 3.5 which means that the fragmentary primes were compatible with many lexical hypotheses. The Baseline, Full Prime and the Baseline, Partial Prime conditions use a full word (e.g., [daqiiqun] *accurate*) and a fragment of it (e.g., [daqii]) as respective baseline primes for the +WP, Full Prime and the +WP, Partial Prime conditions.

To provide a phonological control condition which assesses form overlap from sequence onset, the second set of 24 targets was paired with the same set of prime words as illustrated in **Table 3**. The +Phon, Full Prime condition with prime-target pairs like [wuquuʕun]-[tubuudila] *happening-to be exchanged*, assesses the extent to which pure phonological overlap in the sense of sharing a number of vowel segments that do not make up the same morpheme can be facilitatory. The amount of vocalic overlap in this condition is the same as that in the +WP, Full Prime condition. However, in the +WP, Full Prime condition, the shared vowels make up the same nominal morpheme with singular meaning in the prime and target, while in the +Phon, Full Prime condition, the vowels in the target are in the context of a verb and convey a passive perfective meaning. In the +Phon, Partial Prime condition, the fragment [wuquu] excised from the full prime [wuquuʕun] *happening* is paired with the target [tubuudila]. The question here is whether partial phonological overlap can trigger access to the related target. Finally the target [tubuudila] is paired with the unrelated full prime [daqiiqun] *accurate* in the Baseline, Full Prime condition, and by its fragment [daqii] in the Baseline, Partial Prime condition. In this experiment, prime and target pairs shared on average 62.87% of their phonemes in Condition 1a, 34.53% in condition 1b, 49.80% in condition 2a, and 43.32% in condition 2b.

The numbers of word-word and word-nonword fillers used were similar to those in Experiment 1. The prime-target relatedness proportion was kept at 20%. Additionally, 40 practice trials that were representative of the experimental trials were selected. Four counterbalanced experimental lists were constructed each consisting of 280 pairs.

Procedure

This was identical to the procedure for Experiment 1.

RESULTS AND DISCUSSION

The data for 2 participants were rejected because of high error rates (above 12%). Error trials were excluded (3.51%). Cut-offs were set at 3000 ms and below 100 ms and excluded only 0.05% of the data. The remaining data were inverse transformed to reduce the effects of outliers. Item and participant means were then calculated (see **Table 4**) and analyzed as before using the variables

Table 4 | Reaction times, (standard deviations), amount of priming, and %error rates for the target preceded by test and baseline primes in the different conditions of experiment 2.

Condition	Test	Baseline	Priming	Error in Test	Error in Baseline
+WP, Full Prime	546 (46)	579 (74)	33	4.26	3.49
+WP, Partial Prime	595 (67)	598 (75)	03	3.29	3.49
+Phon, Full Prime	604 (84)	585 (81)	-19	4.26	2.71
+Phon, Partial Prime	593 (79)	602 (81)	9	4.26	2.33

Conditions (morphology vs. phonology), Prime Type (related vs. unrelated), Prime Length (full prime vs. partial prime). A dummy variable representing either the participants grouping in the allocation of subjects to experimental list or the test item grouping in the allocation of items to lists, was included to reduce the estimate of random variation.

The main effect of condition was significant [$F_{(1, 85)} = 10.89$, $p < 0.001$; $F_{(2, 47)} = 3.52$, $p < 0.05$] as was that of Prime Length [$F_{(1, 85)} = 5.81$, $p < 0.05$; $F_{(2, 47)} = 4.31$]. The main effect of Prime Type was not significant [F_1 and $F_2 < 1$]. Condition interacted significantly with Prime Length [$F_{(1, 85)} = 17.55$, $p < 0.001$; $F_{(2, 47)} = 6.95$, $p < 0.05$] reflecting the fact that the partial prime and the full prime had opposite effects in the morphological and phonological conditions. The critical three-way interaction between Condition, Prime Length and Prime Type [$F_{(1, 85)} = 15.16$, $p < 0.001$; $F_{(2, 47)} = 5.27$, $p < 0.05$] indicated that priming was not constant across conditions. Further planned comparisons using Bonferroni protection levels at 0.05, showed that (a) priming effects were significant only in the [+WP, Full Prime] condition, [$F_{(1, 85)} = 6.74$, $p < 0.05$; $F_{(2, 23)} = 5.55$, $p < 0.05$] and the [+Phon, Partial Prime] condition [$F_{(1, 85)} = 5.24$, $p < 0.05$; $F_{(2, 23)} = 5.35$, $p < 0.05$], and (b) that these two conditions differed significantly from the [+WP, Partial Prime] and the [+Phon, Full Prime] conditions, but not from each other [$F_1 < 1$, $F_2 < 1$]. Similar analyses of the error data revealed no significant main effects or interactions.

Finally, to check on the possible contribution of the relevant stimulus properties (duration of the prime, and degree of phonemic overlap between prime and target), these two variable were centered and used in a separate stepwise regression analysis to determine the extent to which they modulate priming. Neither variable significantly predicted priming [prime duration: $R^2 = 0.00$, $F_{(1, 94)} = 0.36$, $p = 0.84$; phonemic overlap: $R^2 = 0.02$, $F_{(1, 94)} = 2.54$, $p = 0.11$].

These results are in keeping with the predictions developed within the CV-hypothesis. Priming by full word patterns is expected on this account since this is a structural unit that allows the drawing of generalization about the phonological structure and morpho-syntactic function of words. Also consistent with this view is the absence of facilitation by partial primes and by phonologically related primes both full and partial. In the case of partial primes, not enough information is provided about the phonological structure of the word, let alone its morpho-syntactic function, so no specific word pattern can be extracted and no savings can be made on the processing of the target. The full

phonological prime is treated as a competitor since it is comprised of a root and a pattern that are different from those of the target (Frauenfelder et al., 2001).

GENERAL DISCUSSION

The results of this study are consistent with the claims of the CV-hypothesis that consonants and vowels need to be segregated both in terms of representation and processing. More importantly, this study extends the CV-hypothesis in a significant way. In terms of processing dynamics this study shows that there is a differential time course at which information about consonants and vowels is mapped onto internal representations. Specifically, partial information about consonants (roots) is continuously used to generate lexical hypotheses, while partial information about vowels (word patterns) is ineffective in accessing the lexicon; only full information about the word pattern provides a basis for lexical access.

The differential processing mechanisms engaged by consonants and vowels suggest that access to information conveyed by consonants (roots) precedes access to information conveyed by vowels (patterns). This is consistent with what we know from masked priming and neuro-physiological research which clearly suggest that the lexical access process in Arabic is oriented toward the consonants (roots) (Boudelaa and Marslen-Wilson, 2005; Boudelaa et al., 2009). These differential processing dynamics arguably originate in the distinct function that consonants and vowels fulfill (McCarthy, 1981; Nespor and Vogel, 1986). Across languages, consonants convey constraining lexical information, and Semitic languages are a clear case where lexical meaning is the domain of the consonantal root. Since the task of the listener is to use the speech input to access meaning, a successful heuristic in Semitic languages is to rely on the consonants of the root. Vowels on the other hand primarily carry phonological and morpho-syntactic information. Apparently this information does not become available unless the full vowel pattern is heard.

It could be argued that the different processing time courses for consonants and vowels in Arabic stems simply from the fact that there are almost 5 times as many consonants in this language (28 consonants) as there are vowels (6 vowels). Based on this simple fact, individual consonants should narrow down the range of possible words more than vowels, and consequently the lexical access process would be oriented toward consonants (roots). However, this distributional bias in favor of the consonants should in fact result in their taking more time to recognize. If we assume, for the sake of argument, that the probability of a given consonant in the language is 1/28 and the probability of a given vowel is 1/6, the chances of making a correct guess regarding the identity of particular segment are much higher for vowels than consonants. Thus, the statistical distribution of the sounds of the language alone, leads us to expect vowels to be easier to recognize and more readily to project onto internal lexical representations. This is obviously not the case; consequently a statistical explanation of the present results is not viable.

In terms of the architecture of the lexicon, the results establish that vowels and consonants are represented independently, not at peripheral levels of modality specific representation as is

the case with Indo-European languages (Caramazza et al., 2000; Carreiras et al., 2007, 2009), but more significantly at higher levels of the language processing system since this is the only site where the processing of an auditory prime can have any processing consequences for a visual target (Marslen-Wilson et al., 1994). Maintaining such distinct representations for consonants and vowels at higher levels of the system makes sense in the context of Semitic languages given the important functional implications such a distinction has for various domains of knowledge such as semantics and morphology and syntax.

The differences between vowels and consonants at the processing and representational levels in this study are at least in part due to the morphemic status that these elements play in the language. Consonants and vowels in Arabic are not simply distinct classes of phonemes, but morphemes with overt implications for semantic meaning and morpho-syntactic functions. However, a potential problem with this interpretation is that although roots are exclusively made up of consonants and word patterns are essentially made up of vowels, several Arabic word patterns feature a subset of consonants along with the vowels (e.g., {ma- -a-} *place noun*, or {muta-aa-i-} *agent noun*). So what processing mechanism applies to consonants that are part of the word pattern? How does the system determine that the consonant /m/ for instance is part of the word pattern in the word [masrah] *theater*, but part of the root in the word [malak] *king*? Pilot data using words starting with a consonant that is either part of the pattern as in [masrah] *theater* or part of the root as in [malak] *king* suggest that the language processor initially treats the sound /m/ similarly in the two words. It uses it in combination with other consonants in the input to access the lexicon. The words [masrah] *theater* and [malak] *king* seem to activate the same set of candidates initially, but as more input is accumulated, different sets of candidates become more viable and get activated accordingly (Magnuson et al., 2007; Jesse and Massaro, 2010). This means that consonants are processed continuously and vowels discontinuously until the correct components of the word, that is the correct root and word pattern, are extracted.

For any model to accommodate the effects of continuous mapping of consonants and discontinuous mapping of vowels it must distinguish between these two elements in terms of representation and processing mechanisms as suggested by the CV-hypothesis, and elaborated here. Independently represented consonants (i.e., roots) act as direct targets for speech input in order to support the continuous mapping necessary for immediate access and efficient communication, while independently represented patterns of vowels will modulate the interpretation of the utterance at later processing stages.

IMPLICATIONS FOR EDUCATION AND REHABILITATION

The dissociation between consonants (roots) and vowels (word patterns) in the context of Arabic has important potential consequences for language practitioners in neuro-rehabilitation and neuro-education. Educators can make curricular changes based on this and similar studies by designing teaching materials where the distinction between the consonants (roots) and vowels (word patterns) and their functions is brought to the fore such that the language learner can build an awareness of these elements.

This awareness can play an important role in helping not only unimpaired learners, but also children whose learning disabilities stem from deficiencies in metalinguistic skills (Bialystok et al., 2014; Tong et al., 2014). In this respect, a recent study by Kim et al. (2013) suggests that awareness of various linguistic domains such as phonology, orthography and morphology provide an effective predictor of reading abilities. More relevantly, morphological awareness generally defined as the child's conscious ability to reflect on and manipulate the structure of his/her language (Carlisle, 1995, p. 194) has been shown to be strongly associated with the child's reading development in languages such as English (Carlisle, 2000), French (Casalis and Louis-Alexandre, 2000) and Chinese (Ku and Anderson, 2003). The present study suggests that in the end-state mental lexicon of Arabic speakers consonants and vowels have different roles by virtue of the different morphemes with which they are typically associated. Language practitioners can capitalize on this finding and develop research informed syllabi that promote awareness of consonants and vowels as different morphemes. Consciously knowing how morphemes fit together and what kind of information they convey should facilitate the acquisition of reading as well the reading of novel words. A child who has a good grasp of the functional properties of consonants (roots) and vowels (word patterns) in Arabic will be better able to figure out the meaning of items like [haasuub] *computer* and [ʔawlama] *globalize* when he/she first hears them. It will be relatively apparent for this child that the form [haasuub] breaks into the consonantal root {hsb} with the general meaning of *counting* and the vowel pattern {-aa-uu-} with a *singular noun* meaning because the child consciously knows what roots and patterns are and they have experienced the same morphological elements in other contexts such as [hisaab] *counting/calculus*, [mahsuub] *counted* etc. . .

In terms of the rehabilitative implications of the current study, the development of diagnostic test batteries and therapeutic methods should be guided by the present (and earlier) results. This paper offers an account of how Arabic consonants (roots) and vowels (word patterns) are differentially mapped onto internal representations of form and meaning. If such a model is accepted, then the language processing system can malfunction only in certain ways (Bullinaria and Chater, 1995; Pulvermüller et al., 2001; Small, 2004). One of these for instance is that in Arabic consonants and vowels can be selectively impaired and spared. Alternatively, deficits in the processing of the consonants of the root may co-occur with semantic deficits since the consonants convey meaning, whereas deficits in vowel processing should ally themselves with problems at the phonological level. This suggests that an effective aphasia test battery for Arabic needs not only to weight morphology as a domain of knowledge that is distinct from other domains, but it also needs to acknowledge the differential properties of different morphemes (i.e., root consonants and vowel patterns) in the processing and representation of the Arabic language. Failing this, the test may not be able to detect the patterns of selective deficits predicted by the model. In conclusion, the development of test batteries to assess acquired or developmental disorders of Arabic should do so in the context of emerging research findings, such as those reported here, about the specific properties of Arabic as a psycholinguistic system.

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Tactile stimulations and wheel rotation responses: toward augmented lane departure warning systems

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When an on-board system detects a drift of a vehicle to the left or to the right, in what way should the information be delivered to the driver? Car manufacturers have so far neglected relevant results from Experimental Psychology and Cognitive Neuroscience. Here we show that this situation possibly led to the sub-optimal design of a lane departure warning system (AFIL, PSA Peugeot Citroën) implemented in commercially available automobile vehicles. Twenty participants performed a two-choice reaction time task in which they were to respond by clockwise or counter-clockwise wheel-rotations to tactile stimulations of their left or right wrist. They performed poorer when responding counter-clockwise to the right vibration and clockwise to the left vibration (incompatible mapping) than when responding according to the reverse (compatible) mapping. This suggests that AFIL implements the worse (incompatible) mapping for the operators. This effect depended on initial practice with the interface. The present research illustrates how basic approaches in Cognitive Science may benefit to Human Factors Engineering and ultimately improve man-machine interfaces and show how initial learning can affect interference effects.

Keywords: driving, tactile reaction time, stimulus-response compatibility, practice, categorization

INTRODUCTION

Thirty seven percent of all transportation fatalities in the USA are caused by running off from the road (National Highway Traffic Safety Administration: <http://www.nhtsa.gov/NCSA>). To cope with this problem, different major car companies (i.e.; Toyota, Honda, Audi, General Motors, Kia Motors, Nissan, Mercedes-Benz, BMW, Opel, PSA Peugeot Citroën) have developed “lane departure warning systems,” that are mechanisms designed to warn the driver when the vehicle is leaving its lane on freeways and arterial roads. The first function of such systems is to allow the driver to engage correcting movements on the steering wheel.

During the past two decades, basic research in Experimental Psychology and Cognitive Neuroscience has tremendously improved our knowledge of the brain mechanisms involved in action control. These advances are in the public domain and could be used to improve man-machine interactions. However, before practical recommendations can be formulated, predictions derived from theoretical constructs must be submitted to empirical testing. The present paper illustrates the first step of a research process that may ultimately augment lane departure warning system in automobile vehicles.

Stimulus-response compatibility (SRC) is a key factor for designing man-machine interfaces. It refers to the fact that some actions are easier or more difficult than others either because of the particular sets of stimuli and responses that are used or because of the way in which individual stimuli and responses are paired with each other (Kornblum et al., 1990). Hommel et al. proposed a general frame, the theory of event coding (TEC), that explains how stimuli and responses are represented and how these

representations interact to generate SRC (Hommel et al., 2001; Hommel, 2009). At the core of TEC is the notion that produced actions (responses) are represented in terms of their perceptual consequences (stimulus). Responses and stimuli thus share some features in a common representational domain. Representations consist in composite feature codes organized in networks. The more features are shared by stimulus and response representations, the more compatible are the events they refer to. Feature overlap between stimuli and responses representations thus cause SRC effects.

Here, we shall focus to the lane departure warning system implemented in PSA Peugeot Citroën vehicles (AFIL, for “*alerte de franchissement involontaire de ligne*”) and bring empirical evidence that this system is potentially sub-optimal. In this system, the warning consists in a vibration delivered to the driver through the seat on the side of the lane departure. When receiving this warning, the driver must rotate the steering wheel so as to replace the vehicle in its lane (for studies on drivers’ steering reactions to disturbances, see Muto and Wierwille, 1982; Wierwille et al., 1983; Franck et al., 1988). A first caveat of AFIL is to rely exclusively on the tactile modality. Multisensory displays that are based on the latest cognitive neuroscience research findings can capture driver attention significantly more effective than their unimodal (i.e., tactile) counterparts (for a review, see Spence and Ho, 2008). The second, and most critical point, is that AFIL delivers the tactile warning to the driver on the same side as the lane departure: when the vehicle runs off the lane on the left side, the left side of the drivers’ seat is vibrated while when the vehicle runs off the lane on the right side, the right side of the drivers’ seat is vibrated.

Since different studies (Rieger et al., 2005; Sutter, 2007; Müsseler et al., 2008) showed that anticipated effects in external space are prevalent for SRC when using tools, delivering the warning on the side of the body corresponding to the lane departure (proximal reference) rather on the side of the to-be-performed corrective movement (distal reference) seemed a questionable option (for a similar analysis, see Straughn et al., 2009). It must further be stressed that “motor priming,” a recent prototype of device assistance, implements a mapping opposite to that of AFIL: In case of lane departure it delivers small alternating movements to the steering wheel directed toward the road center (Navarro et al., 2007, 2010).

The aim of the present study was to help deciphering the optimal way of delivering the tactile warning to the driver after the on-board system has detected a drift of the vehicle to the left or to the right. To this end, two stimulus-response mappings were contrasted. As AFIL, the first one favored the proximal references: It consisted in responding by a clockwise rotation of the steering wheel to the left-side stimulus and by a counter-clockwise rotation to the right-side stimulus. The second one favored anticipated effects in external space: It consisted in responding by a counter-clockwise rotation of the steering wheel to the left-side stimulus and by a clockwise rotation to the right-side stimulus.

According to TEC, the larger feature overlap between the stimulus and response representations, the larger the difference in performance between the mappings (Hommel, 2009). In AFIL, the lateralized warnings are delivered through the seat which does not insure maximal feature overlap between the stimulus and response representations. One way to augment this overlap so as to maximize the difference in performance between the two possible mappings consists in delivering the tactile warnings to a part of the body's driver directly involved in the steering rotation response. Here, in order to render the hypotheses testable with a limited number of participants, we chose to deliver the warnings to the wrists. This option was further technically easy to implement in a first approach. Future developments could be based on stimulating the palm of the hand through vibrators inserted in the steering wheel.

In driving conditions, any drift of the vehicle is inevitably accompanied with changes in the visual scene. In an attempt to design a situation closer to driving conditions, visual feed-backs relative to the orientation of the steering wheel were provided in the present study. The wheel movements therefore produced visual spatial effects entailing the confounding of stimulus-response compatibility with response-visual effect (feed-back) compatibility, which influence has been demonstrated (Hommel, 1993, 1996; Kunde, 2001; Kunde et al., 2002). For instance, Kunde (2001) demonstrated that keypresses are initiated faster when they trigger visual events in spatially corresponding rather non-corresponding locations. For the present purpose, the covariation of these two variables, which occurs under natural driving conditions, is unproblematic inasmuch it allows one to address the question of how the tactile information should be delivered to the driver and renders the experimental design more realistic.

While the results obtained in an applied study are inconclusive (Beruscha et al., 2010), basic research results (Guiard, 1983; Stins and Michaels, 1997; Proctor et al., 2004; Murchison and

Proctor, 2013), lead us to expect the participant's performance to be better when the subjects responded counter-clockwise to left stimulations and clockwise to right stimulations than when they performed the reverse mapping. Compatibility being to a great extent a matter of learning (Kornblum et al., 1990), a second aim of the present study was to investigate how practicing the alternative mappings may affect the participants' performance.

MATERIALS AND METHODS

PARTICIPANTS

Twenty right-handed participants, 8 women and 12 men, aged 21–62 years (mean: 37, *SD*: 11), and holding a car driving license on average for 17 years (*SD*: 11, range 1–43) volunteered for the experiment. All of them had a normal or corrected-to-normal visual acuity. They were split into two groups of 10, each group comprised 4 women and 6 men. The participants of group 1 were aged 21–61 years (mean: 38, *SD*: 11) and were holding their car driving license on average for 18 years (*SD*: 12, range 1–43). The participants of group 2 were aged 25–62 years (mean: 36, *SD*: 12) and were holding their car driving license on average for 16 years (*SD*: 12, range: 3–42).

TASK

Apparatus and display

The participant was seated comfortably on a chair and was to grip with the two hands a Microsoft® Side winder® steering wheel, 26 cm in diameter, interfaced to a Pentium 4 equipped micro-computer. In front of the participant, behind the steering wheel, a 21 inches computer screen was disposed at eye level. This screen served to display visual feed-backs. The distance between the screen and the participants' eyes was about 70 cm. The stimuli were vibrations (Frequency 108 Hz, Amplitude 0.46 mm, duration 200 ms) applied to the internal part of the two wrists by electrical rotary engines (Deltron Euroind Company, Italy) inserted in cloth braces maintained by Velcro fixations. Responses were clockwise or counter-clockwise rotations of the steering wheel. Visual feedbacks were delivered on the computer screen. They consisted in triangles (Base = 9 cm, Height = 9 cm) of green (RGB = 19, 225, 0), blue (RGB = 0, 0, 255) or red (RGB = 255, 0, 0) color, carrying information detailed below.

Trial

Participants gripped the steering wheel with their hands in a typical driving position: their left and right hands being respectively on the left and right side of the wheel (10:10 grip). Each trial began by positioning the steering wheel in the starting position (between -64° and $+0.64^\circ$ with regard to the vertical). Once this position was reached, a green triangle pointing upward was displayed on the screen.

Provided that the steering wheel was kept in the starting position during 500 ms, a tactile stimulation was delivered either to the left or to the right wrist. The time allowed for the participants to leave the starting position and reach the target position was 800 ms. The target position was reached by rotating the steering wheel, either clockwise or counter-clockwise, depending on the stimulation (see below), to reach a position of $37.2^\circ + 6.4^\circ$ with regard to the vertical. The rotation of the steering wheel

turned-off the green triangle and a blue triangle pointing either to the right, for a clockwise rotation, or to the left, for a counter-clockwise rotation, appeared on the screen. When the steering wheel reached the target position, the blue triangle turned green. If the steering wheel went too far (beyond the target), the green triangle turned red. All triangles were pointing to the right for a clockwise rotation and to the left for a counter-clockwise rotation. Participants had to maintain the steering wheel in the target position during 1000 ms. When they succeeded, the response was correct and an auditory positive feedback was emitted (Windows_XP_Sound_by_default.wav); otherwise, the response was incorrect and an auditory negative feedback was delivered (Windows_XP_Discharged_battery.wav). A response was considered as an error when: (1) participants did not react in time (2) participants did not reach the target in time (3) participants reached the target but did not keep the position (4) participants went in the opposite direction.

INSTRUCTIONS AND MAPPINGS

Instructions were given verbally by the experimenter and emphasized both speed and accuracy. For one mapping, participants were asked to respond by a counter-clockwise rotation to the vibration of the left wrist and by a clockwise rotation to the vibration of the right wrist; for the other mapping, participants were asked to respond by a clockwise rotation to the vibration of their left wrist and by a counter-clockwise rotation to the vibration of their right wrist. Note that these formulations make no verbal reference to a possible lateral coding of the wheel-rotation responses.

DESIGN

The participants participated in four daily sessions. During each session, they performed first 15 warm-up trials and then 5 experimental blocks of 64 trials. The warm-up trials during each session were performed using the same mapping as the following five experimental blocks. Within a block, the two stimuli were equiprobable and delivered according to a pseudo-random sequence. The participants were given a few minutes of rest between each block. Mapping was alternated every other session. Group 1 began by responding counter-clockwise to the left stimulation and clockwise to the right stimulation; Group 2 did the reverse assignment of mapping to session (see **Table 1**).

DATA ANALYSIS

Mean RT was submitted to univariate repeated-measures analysis of variance (ANOVA). The design involved one between-subject factor, group (i.e., mapping sequence, two levels), and two within-subject factors, block of trials (five levels) and session (four levels). Mean error rates of “Side errors” (participants moved the steering wheel in the opposite direction) were arcsine transformed and submitted to analyses of variance with the same design as that used for the RT data. Other incorrect trials included “Late movements” (participants did not reach the target in time: with a movement time between 800 and 1600 ms), and “Stabilization” (participants reach the target but did not keep the position). These trials were also arcsine transformed and submitted to analyses of variance with the same design. Note that percentage data

cannot be tested by parametric tests as their means and variances are closely related. However, the arcsine transform is efficient in stabilizing the variances of these data (Winer, 1970). In a few trials (<1%), participants did not react during the 800 ms following the stimulation; these omissions were judged too few for analysis.

RESULTS

REACTION TIME

Mean RTs are presented in **Figure 1** and **Table 2**. The participants of group 1 “Compatible first” responded by a counter-clockwise rotation to the left vibration and by a clockwise rotation to the right vibration during the first and third sessions while this mapping was performed by the participants of group 2 “Incompatible first” during the second and fourth sessions. Symmetrically, the participants of group 2 “Incompatible first” responded by a clockwise rotation to the left vibration during the first and third sessions while this mapping was performed by the participants of group 1 “Compatible first” during the second and fourth sessions. To test the effect of S-R mapping, we thus compared the first and third sessions together against the second and fourth sessions. This comparison revealed that the effect of session differed as a function of the group of participants [$F_{(1, 18)} = 9.43$, $p = 0.01$, $\eta^2 = 0.01$].

Conducted separately for each group, this comparison revealed that the participants of group 1 “Compatible first” reacted faster when they responded by a counter-clockwise rotation to left stimulation and a clockwise rotation to right stimulation than when they performed the reverse mapping [$F_{(1, 9)} = 7.98$, $p = 0.02$, $\eta^2 = 0.02$]. Therefore, the former mapping was more compatible than the later one. Further comparisons allowed one to refine this analysis. For group 1 “Compatible first,” there was no significant difference in RT between the first and third sessions (compatible mapping) nor between the second and fourth sessions (incompatible mapping, $ps > 0.10$). In other words, the effect of S-R mapping was all-or-none: It was unaffected by the repetition of mapping sessions (see **Figure 1**).

Contrary to those of group 1 “Compatible first,” RTs of participants of group 2 “Incompatible first” who practiced the incompatible mapping during the first and third sessions were not

Table 1 | Assignment of mappings to sessions for the two participants’ groups.

	Group 1	Group 2
Session 1	Left S / counter-clockwise R Right S / clockwise R	Left S / clockwise R Right S / counter-clockwise R
Session 2	Left S / clockwise R Right S / counter clockwise R	Left S / counter-clockwise R Right S / clockwise R
Session 3	Left S / counter-clockwise R Right S / clockwise R	Left S / clockwise R Right S / counter-clockwise R
Session 4	Left S / clockwise R Right S / counter clockwise R	Left S / counter-clockwise R Right S / clockwise R

S, stimulus; R, response.

Table 2 | Mean reaction time and percentage of incorrect trials sorted by type (Side errors, Late, Stabilization, Omission) for each group (Group 1 “Compatible first,” Group 2 “Incompatible first”) and for each condition (session, block of trials).

Session (S)	Block	Group 1 “Compatible first” (G1)						Group 2 “Incompatible first” (G2)					
		RT (<i>M</i> ; <i>SD</i>)		Side	Late	Stab.	Omis.	RT (<i>M</i> ; <i>SD</i>)		Side	Late	Stab.	Omis.
S1	1	378	39	1.4	2.5	7.0	0.0	409	78	7.2	1.9	7.3	0.6
	2	359	39	1.9	1.2	7.5	0.3	394	72	5.3	4.8	6.9	0.6
	3	359	41	2.7	1.2	6.2	0.0	386	64	3.8	1.6	3.8	0.2
	4	354	42	2.2	0.6	6.1	0.2	381	58	3.6	2.7	4.5	0.0
	5	354	39	1.9	1.9	5.9	0.0	382	62	2.5	2.3	3.9	1.2
Mean		361	39	2.1	1.5	6.5	0.0	390	66	7.2	1.9	7.3	0.6
S2	1	380	56	5.2	1.7	5.9	0.5	384	67	3.1	0.6	3.0	0.8
	2	378	48	4.2	0.9	5.0	0.0	390	74	1.9	0.8	2.7	0.6
	3	382	49	3.9	1.4	3.4	0.3	377	62	0.9	1.1	1.2	0.2
	4	377	55	2.5	0.8	4.4	0.2	375	67	1.1	1.2	0.9	0.5
	5	376	48	2.8	1.4	5.3	0.2	374	62	1.9	0.5	2.5	0.3
Mean		379	50	5.2	1.7	5.9	0.5	380	66	3.1	0.6	3.0	0.8
S3	1	360	26	0.8	0.2	4.8	0.0	383	66	3.1	1.1	2.2	0.0
	2	348	30	1.2	0.8	3.9	0.0	388	67	0.8	0.3	1.6	0.0
	3	348	27	1.9	0.3	3.6	0.2	387	64	1.9	0.6	1.1	0.3
	4	346	29	0.6	0.5	3.4	0.0	379	62	1.1	0.5	1.7	0.8
	5	343	32	1.4	0.5	3.3	2.0	380	58	0.9	0.3	1.6	0.2
Mean		349	28	0.8	0.2	4.8	0.0	383	63	3.1	1.1	2.2	0.0
S4	1	372	36	2.7	0.3	4.5	0.0	384	63	1.6	0.5	2.2	0.8
	2	373	36	2.2	0.0	3.1	0.0	389	71	0.9	0.5	2.0	1.1
	3	375	38	3.8	0.3	5.2	0.0	378	64	1.4	0.2	0.5	0.2
	4	372	35	3.6	0.9	4.8	0.2	384	67	1.1	0.3	2.7	0.9
	5	367	35	3.3	1.1	5.3	0.0	374	63	1.2	0.6	0.8	0.2
Mean		372	35	2.7	0.3	4.5	0.0	382	65	1.6	0.5	2.2	0.8
Mean (S1, S3)		355	31	1.4	0.8	5.7	0.0	387	64	5.2	1.5	4.8	0.3
Mean (S2, S4)		375	41	4.0	1.0	5.2	0.3	381	65	2.4	0.6	2.6	0.8

significantly affected by the S-R mapping ($p > 0.10$). In addition, a direct comparison of the two groups revealed that mapping sequence did not significantly influence RT for the second and fourth sessions ($p > 0.10$), indicating that participants of group 2 “Incompatible first” in the compatible mapping reacted as slow as participants of group 1 “Compatible first” in the incompatible mapping. The two mappings appeared thus equally incompatible for the participants of group 2 “Incompatible first” (see **Figure 1**).

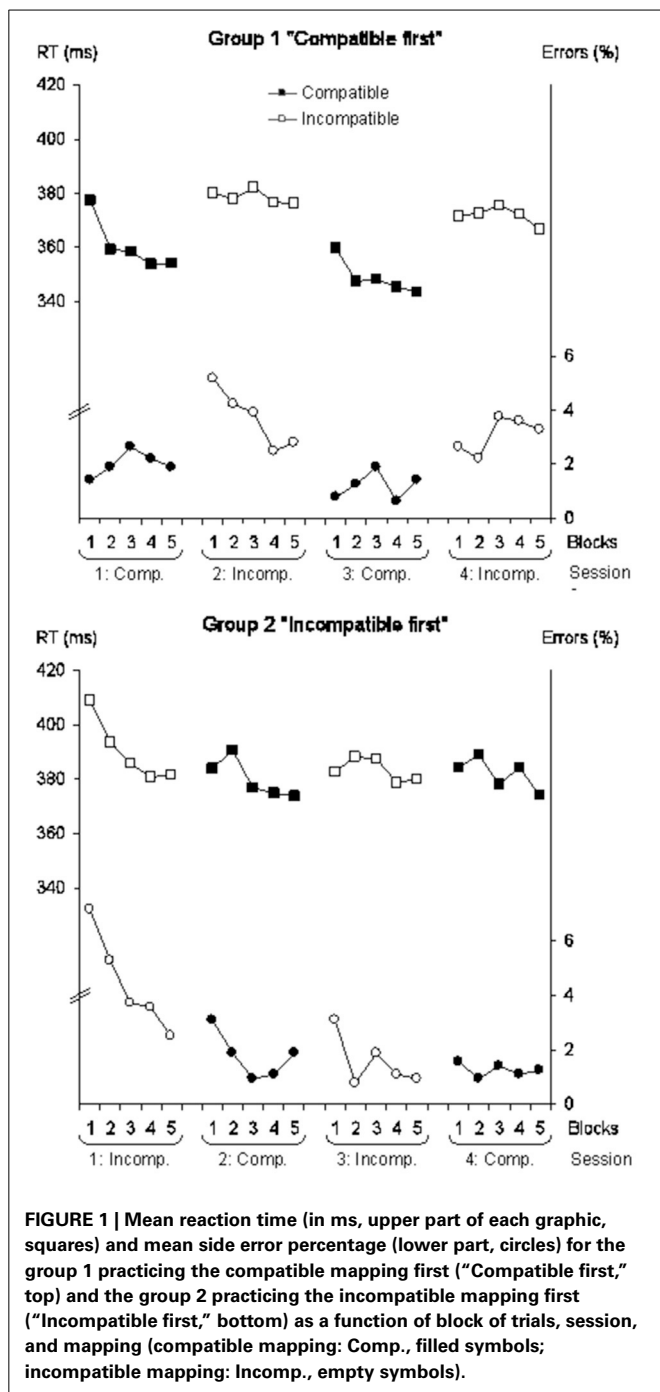
In an attempt to better characterize the effects on mean RT, a distribution analysis was also performed. As the mapping effects can be partially confounded with those of with-session practice in the first two sessions, we focused the distributional analysis on the last two sessions. Individual RT distributions were “Vincetized” (Ratcliff, 1979); each individual RT distribution was binned in ten classes of equal size (same number of trials) and the mean of each bin was computed. **Figure 2** presents the averaging of these individual RT distributions. It can be seen that participants of group 1 “Compatible first” consistently had a better performance

for the compatible than for the incompatible mapping across all distribution deciles, contrary to the participants of group 2 “Incompatible first.”

ERROR

Side errors

The side errors (participants moved the steering wheel in the opposite direction) represent 2.4% of the trials; mean side error percentages are presented in **Figure 1**. We compared the first and third sessions together against the second and fourth sessions for each group of participants, as for the reaction time. This comparison showed that the effect of session differed as a function of the group of participants [$F_{(1, 18)} = 22.34, p < 0.01, \eta^2 = 0.08$]. This comparison conducted separately for each group revealed that, contrary to the participants of group 2 “Incompatible first,” the participants of group 1 “Compatible first” made significantly less errors when they were required to perform an counter-clockwise rotation when stimulated to the left wrist and



a clockwise rotation when stimulated to the right wrist than when they were required to perform the reverse mapping [$F_{(1, 9)} = 28.69$, $p < 0.01$, $\eta^2 = 0.07$; group 2: $F_{(1, 9)} = 4.41$, $p = 0.07$]. The former mapping was found more compatible than the latter one, which parallels the results on the reaction time (see Figure 1).

Further comparisons showed that, for group 1, there was no difference neither between the first and third sessions (compatible mapping) nor between the second and fourth sessions (incompatible mapping; $ps > 0.10$). For group 2, there was no

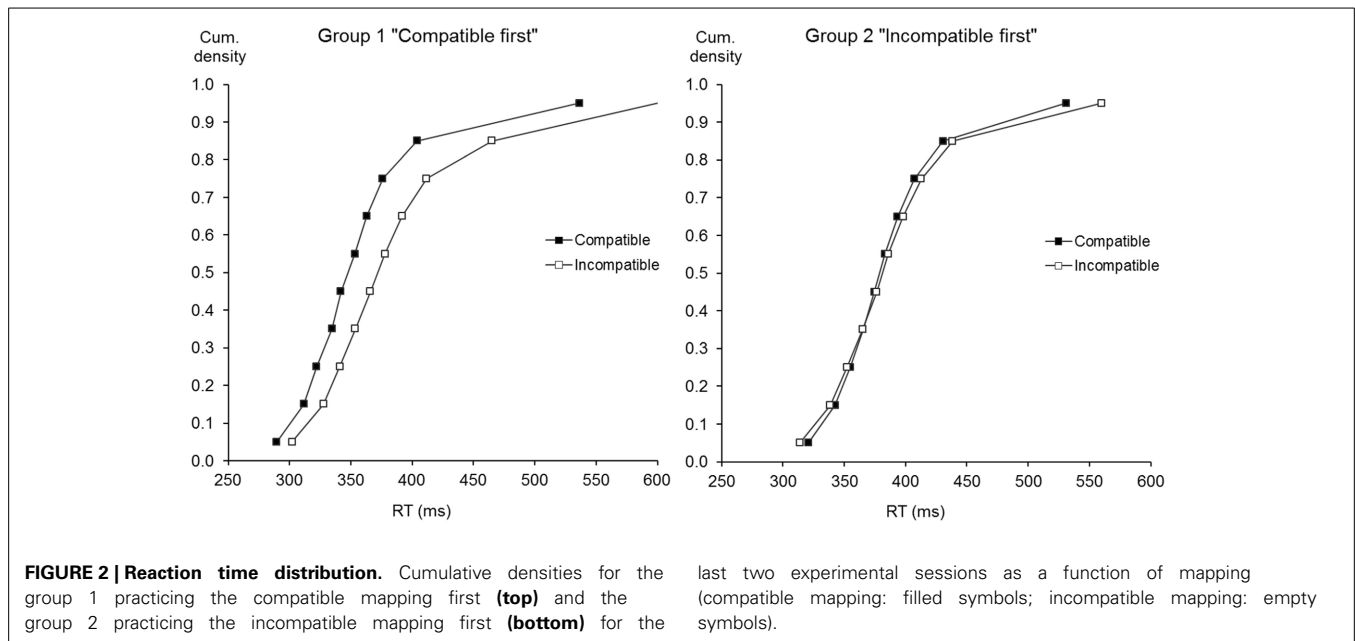
difference between the second and fourth sessions ($p > 0.10$) but a significant difference between the first and third sessions [$F_{(1, 9)} = 8.60$, $p = 0.02$]. This pattern of results on side errors indicate an effect of S-R compatibility for the group 1 but an effect of between-session practice for the group 2 (see Figure 1).

Other incorrect trials

Late movements (participants did not reach the target in time: with a movement time between 800 and 1600 ms), Stabilization (participants reach the target but did not keep the position), and Omissions (participants did not react during the 800 ms following the stimulation) represent 3.8, 1.0, and 0.3% of the trials, respectively (Table 1). There were no significant effect of S-R mapping on "Late movements" and "Stabilization" trials, as assessed by the comparison between the first and third sessions together against the second and fourth sessions ($ps > 0.10$).

DISCUSSION

The participants of group 1, who initially responded counter-clockwise to the left vibration and clockwise to the right vibration, displayed a clear effect of compatibility on both RT and side error rate. This mapping remained more compatible than its alternative throughout the experiment. The participants of group 2, who first responded clockwise to the left vibration and counter-clockwise to the right vibration displayed no hint of compatibility effect. Thus, depending on initial practice, the mapping selected for AFIL (PSA Peugeot Citroën) was either neutral or detrimental to the participants' performance. A first implication of these findings is that it seems preferable to design systems requiring the operators to respond to left tactile stimulations by counter-clockwise wheel rotations and to right tactile stimulation by clockwise rotations. It can be stressed that this outcome is in line with predictions derived from TEC (Hommel, 2009) which posits that anticipated effects in external space (distal references) as opposed to body-centered representations (proximal references) are prevalent for SRC. One may object that the present result are not directly predictive of the drivers' behavior when using AFIL because, with this system, the lateralized tactile stimulations are delivered to the thighs rather than to the wrists. In a first approach, stimulating the wrists was intended to augment feature overlap between the stimuli and response sets in order to maximize the effect of mapping (Hommel, 2009) and allow conclusions from a limited sample of participants. The effect of this methodological choice should be directly addressed in forthcoming experiments, by delivering the warnings through the participants' seat. The hand position (crossed or uncrossed) could also play a role by modulating the compatibility between the tactile stimulation and the wheel-rotation responses. While hand placement plays a role for estimating the temporal order of successive tactile stimuli delivered at very brief intervals (Yamamoto and Kitazawa, 2001), previous research allows clear expectations relative to its influence on SRC. Since anticipated effects in external space are prevalent when using tools (Rieger et al., 2005; Sutter, 2007; Müsseler et al., 2008), best warning should probably be delivered in the direction of the to be performed rotation, irrespective of hand placement on the steering-wheel. The implications of the present results for designing on-board systems



should further be confirmed in more realistic driving conditions (in driving simulators and in real traffic circumstances). This may work when the driver hold a steering wheel with both hands in the horizontal position, i.e., the right hand in the position of 90-degree clockwise from the top and the left hand in the position of 90-degree counterclockwise. This might be a natural position of hands when holding the steering. But, we must notes that there are wide variation in the position of hand. When turning a left bent, the right hand can be in the top of wheel and the left hand in the bottom of wheel. Even, the right hand can be more left and the left hand can be more right when the driver crosses both hands.

Another important issue for future research is to combine the tactile stimulation with an information relative to the vehicle position that is often delivered in motorways: A rumble strip on the lane marker produces a vibration when the car is running off from the lane. The relationship between the tactile stimulus and this vibration can be formalized in terms of stimulus-stimulus compatibility (SSC, Fitts, 1954). Another instance of SSC likely interfered with SRC in an experiment conducted in a driving simulator by Straughn et al. (2009). These authors examined the effect of compatibility between steering responses and accessory left/right tactile signals delivered after visual imperative signals. In this task, the compatibility between the accessory and the response (SRC) and the compatibility between the imperative signal and the accessory (SSC) covaried in opposition. The delay between the imperative signal and the accessory signal was also manipulated. For short delays, the left accessory—counter clockwise response / right accessory clockwise response led to the best level of performance. In contrast, for long delays, the alternative mapping led to the best performance level. While disentangling the respective effects of the two compatibility relationships is beyond the scope of the present paper, it must be stressed that for the short delay condition that is the closest to the present

experiment, the effect of accessory-response compatibility corresponds to the SRC effects evidenced in the present study. The inversion of this effect for long delays shows that the relationship between SRC and SSC should be addressed before firm conclusions relative to the use of onboard lane departure systems can be reached (for an application of this notion in ergonomics, see Akamatsu et al., 1995).

The efficiency of vibrations were compared to that of the “motor priming” device (Navarro et al., 2007) that prompts drivers to take action by means of small asymmetric oscillations (Navarro et al., 2010) producing either a clockwise or a counter-clockwise rotation of the steering wheel. Motor priming implements a compatible mapping: a clockwise rotation calls for a clockwise response and a counter-clockwise rotation counter-clockwise response (toward the road center). Surprisingly, the authors tested motor priming against left and right tactile vibrations incompatibly mapped with the steering response: the left vibration was to be responded to by a clockwise movement and the right vibration by a counter-clockwise movement (toward the lane departure). The present results suggest that motor priming would be better tested against a compatible vibrotactile-motor mapping.

A final comment relative to automotive ergonomics is in order. So far, lateral control assisting devices can be split in two categories: lane departure warning system and lane keeping assistance systems. Lane departure warning systems, such as that tested in the present study, simply inform the driver that the vehicle is in an unsafe lane position. Lane Keeping assistance systems actively intervene on the steering wheel. For instance, torque can be continuously applied to the steering wheel to help the driver to keep close to the center of the lane. Future developments based on motor priming concept (Navarro et al., 2007) may combine the two types of systems. This can be done by taking advantage of the torque already installed for power

steering and may render obsolete systems based on auditory or vibratory warnings (for a recent review see Beruscha et al., 2011).

We suggest that instead of considering from the beginning complex near-to-real man-machine interactions, Human Engineering starts from facts from basic research even established in unpoverished experimental conditions. Applied research could then elaborate validation processes for testing the relevance of these facts under increasing complexity situations progressively approaching real-life conditions. This issue is of societal relevance for such validation processes may hopefully save human lives, in particular in transportations.

The list-rule model of SRC (Hasbroucq et al., 1990) is a functional model compatible with the representational component of TEC (Hommel, 2009). It provides a simple systematic articulation between stimulus-response match and the effect of mapping. Functionally, like other instances of SRC proper (e.g., Fitts and Deininger, 1954), the present effect can be attributed to a central response selection process which transforms the stimulus categorized by preceding identification processes into an abstract response code that is subsequently transmitted to motor stages of information processing (Proctor and Reeve, 1990), a notion that is supported by data from single cell recordings in behaving monkeys (Riehle et al., 1997; Mouret and Hasbroucq, 2000). According to the list-rule model, when the stimuli and responses sets do not match, they are differently categorized and the only response selection procedure available to the subject consists of scanning the memorized list of the individual stimulus-response pairs that have been defined in the instructions as correct. In this case, no effect of mapping is expected. If, alternatively, the stimulus and response sets match, the subjects do commonly categorize the stimuli and responses and the possibility of an algorithmically governed response selection can emerge. In particular, when the mapping associates the counter-clockwise response to the left stimulus and the clockwise response to the right stimulus, the common categorization of stimuli and response in lateral terms allows the subjects to select their responses by applying the identity transformation.

We shall now consider the implications of the effect of mapping sequence for this model and for man-machine interfaces. Indeed, only the participants of group 1 exhibited an effect of SRC. According to the list-rule model, these participants initially completed a mapping fitting their *a priori* left-right categorization of the stimulus and response sets. A difference in response selection procedure across sessions accounts for the emergence of an effect of SRC in this group. In sessions one and three, the subjects of group 1 applied the identity transformation while, in sessions two and four, this simple rule was no more available. They were thus to select their responses either by applying the inverted transformation to the stimuli categorized in left and right terms or to scan the memorized list of the alternative stimulus-response pairs: left / clockwise, right / counter-clockwise. In contrast, the subjects of group 2 exhibited no effect of compatibility. In terms of the list-rule model, these subjects initially completed a mapping that did not fit their *a priori* left-right categorization and they resorted to the same response selection procedure across experimental sessions. Admittedly, for the reason evoked above,

in sessions one and three, the subjects of group 2 scanned the memorized list of the two alternative stimulus-response pairs. Surprisingly, their performance did not improve in sessions two and four during which the stimuli and responses could perfectly be categorized in lateral terms in order to apply the identity rule. The absence of performance improvement in these sessions suggests that the scanning procedure implemented in session one and three overrode the available common categorization of the stimulus and response sets, thereby preventing the use of the identity transformation which would have made emerged an SRC effect comparable to that observed for group 1.

These findings suggest that the categorization processes depend not only on the pre-experimental background of the subjects but also on the mapping according to which the experimental task is initially performed. Indeed, our results indicate that pre-experimental background and experimental practice interact, thereby determining the categorization operated by the subjects throughout the experiment. From a human factors perspective, an implication of this finding is that the first experience with an interface may be critical: it can determine the subsequent performance of individuals with this interface. Taking compatibility factors into account should thus help designing the most favorable learning procedure when operators are confronted with new man-machine interfaces. Since industrial and transportation contexts incur serious potential hazards, this may have strong societal implications. Since optimal man-machine interactions are conditioned by the compatibility of the mapping initially prescribed on a given interface, operators should learn the most compatible mapping. To this aim, the relative compatibility of all possible mappings should be assessed so as learning procedures favor the most compatible one. Finally, in complex situations, determining which associations will be congruent and which ones will not cannot rely on naïve judgments (Payne, 1995; Vu and Proctor, 2003; Hoffmann, 2010) but must be evaluated experimentally. It is noteworthy that such evaluations must take into account learning effects.

With respect to SRC research, there is not much evidence in the literature that learning has pronounced effects on SRC (e.g., Brebner, 1973; Dutta and Proctor, 1992) and the present effect of practice is not easy to reconcile with the functional distinction between long-term and short-term stimulus-response associations advocated by Barber and O'Leary (1997). Recent studies show, however, that preliminary practice of incompatible associations in tasks where the stimulus location is relevant does modify the effect of the irrelevant stimulus location in subsequent tasks (e.g., Tagliabue et al., 2000; Proctor et al., 2003). The present results demonstrate comparable transfer effects when the stimulus location of the subsequent task is relevant.

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