

Neurofeedback as a potential treatment for stuttering

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Abstract

The purpose of this study was to start exploring the feasibility of neurofeedback training based on electroencephalography (EEG) data as a potential treatment for persistent developmental stuttering (PDS). In this preliminary study, part of a larger project, visual feedback based on each participant's slow cortical potentials (SCPs) was integrated in a brain-controlled video game during 12 training sessions distributed over a 6-week period. For each session, behavioral data regarding game performance and pre- and post-training stuttering frequency were collected together with the EEG data. Two of three adult participants with PDS showed some evidence of learning in game performance and/or EEG data, and these two participants also showed limited—and inconsistent—changes in stuttering frequency during oral reading (but not conversation). One participant stuttered less immediately after each training session as compared with prior to each training session, in the absence of any improvement across the sessions. The other participant's post-training stuttering frequency showed a small, gradual reduction across the sessions. The third participant showed no evidence of learning and no trend toward a change in stuttering frequency. These initial pilot data warrant further investigation of (a) SCP-based neurofeedback protocols with more participants and over a longer period of time and (b) training protocols based on different aspects of brain activity.

Neurofeedback as a potential treatment for stuttering

Stuttering is a disorder of speech fluency characterized by involuntary repetitions of sounds, syllables or multisyllabic words, audible or inaudible prolongations of sounds, and/or broken words. In addition to the obvious communication difficulties resulting from these primary characteristics, the disorder often has a strong negative impact on the quality of life for many affected individuals (Bloodstein & Ratner, 2008; Craig et al., 2009). Individuals who stutter report negative emotions (e.g., shame, fear, embarrassment) and stigmatization (Hugh-Jones & Smith, 1999) that affect communication attitudes (Blood et al., 2007; De Nil & Brutten, 1991), self-esteem (Watson 1995), academic achievement (Williams et al., 1969), and interpersonal relationships (Linn & Caruso, 1998). Given that the prevalence of stuttering is approximately 1% regardless of language or culture, as many as 3.5 million individuals stutter in the United States alone (Bloodstein & Ratner, 2008). Hence, the need for an in-depth understanding of the neurobiological bases of the disorder and for the availability of effective, evidence-based treatment approaches cannot be overemphasized.

Most existing treatments for stuttering are behavioral in nature, focusing on speech techniques that are used to modify the stuttering moments (e.g., slowing down and reducing muscular tension during the stuttering itself) or to avoid stuttering moments as much as possible (e.g., using an overall slower speaking rate, easy voice onsets). Unfortunately, these existing treatments are a-theoretical, and many are prone to relapse (Boberg, 1981; Craig, 1998; Jones et al., 2008). In fact, it has been estimated that the relapse rate of existing treatments is 50-70% (Silverman, 1992). Thus, the development

of novel clinical management approaches that yield lasting improvements is of crucial importance.

The initiation of such a clinical paradigm shift, however, requires a strong foundation in contemporary insights from both basic and clinical neuroscience. Although one hypothesis of aberrant hemispheric lateralization of brain activity in individuals who stutter had already been proposed more than eight decades ago (Orton, 1927; Travis, 1931), the more recent development of various neuroimaging techniques has caused new knowledge to accumulate at a particularly fast rate.

Structural neuroimaging studies in both children and adults have shown reduced integrity of the white matter underlying pre-motor and motor speech areas in stuttering vs. nonstuttering individuals (Chang, Horwitz, Ostuni, Reynolds, & Ludlow, 2011; Sommer, Koch, Paulus, Weiller, & Buchel, 2002) as well as more general differences in grey and white matter volume (Beal, Gracco, Lafaille, & De Nil, 2007). Functional neuroimaging studies have revealed numerous differences in stuttering speakers as compared with nonstuttering speakers. One consistent finding is reduced activation of the left hemisphere and increased activation of the right hemisphere, resulting in an absence or reversal of the normal left hemisphere lateralization for speech production (among many others, De Nil, Kroll, & Houle, 2001; Fox et al., 1996). A second finding that has been replicated in several studies is an overall over-activation of motor and premotor cerebral and cerebellar areas (De Nil et al., 2001; Fox et al., 1996, 2000). A third finding of interest is that stuttering subjects fail to activate, or even deactivate (relative to rest), the auditory cortex during speech production (De Nil et al., 2003; Fox et al., 1996, 2000).

Based on these findings, as well as those from numerous other publications, it is reasonable to conclude that stuttering is associated with anatomical and functional deficits in an extensive neural network involved in sensory and motor aspects of speech motor planning and execution. This is consistent with many of our own laboratory's behavioral or psychophysical results suggesting that both children and adults who stutter perform more poorly than their nonstuttering peers on tasks of sensorimotor integration and sensorimotor learning (Daliri & Max, submitted; Daliri, Prokopenko, & Max, 2013; Daliri, Prokopenko, Flanagan, & Max, submitted).

Of particular importance here, however, is the fact that the observed abnormalities in stuttering subjects' brain activation are amenable to modification, presumably as a result of neural plasticity. Indeed, at least some atypical activations have been found to normalize (i.e., become more similar to those of nonstuttering subjects) in fluency-enhancing conditions or after clinical treatment. For example, when Fox et al. (1996, 2000) found that their stuttering subjects showed under-activation of the auditory cortex during speech production, they also observed that this auditory cortical activation became more similar to that of the control group when fluent speech was induced with a technique known as choral reading (whereby an examiner reads out loud together with the stuttering subject; this technique temporarily enhances fluency for many individuals who stutter, but has no therapeutic applications given that the effect does not generalize after the examiner stops reading). Similarly, De Nil et al. (2003) reported that after speech therapy, as compared with prior to the therapy, stuttering subjects showed changes toward a more typical left hemisphere lateralization of brain activation. *Here, we*

hypothesize that direct modification of brain activity through neurofeedback training can lead to reductions in the behavioral speech symptoms of stuttering.

During neurofeedback training, information about temporal, spatial, or spectral aspects of brain activation is provided to the subject in the form of auditory or visual feedback. When provided with such feedback, many (although not all) subjects can learn over time to voluntarily regulate the targeted aspect of their brain activation. A large body of literature on neurofeedback training is already available, and continues to grow at rapid pace. Overall, this literature demonstrates considerable success – i.e., symptom reductions – for applications with a wide variety of neurodevelopmental disorders (e.g., attention deficit hyperactivity disorder – ADHD, Tourette Syndrome) and neurological conditions (e.g., epilepsy). To date, neurofeedback has not been explored as a potential clinical technique for application in the treatment of stuttering.

Although functional magnetic resonance imaging (fMRI) and magnetoencephalography (MEG) also can be used, the most common form of neurofeedback uses surface electrodes on the scalp (i.e., electroencephalography or EEG) and provides the subject with relevant information in a visual display. EEG data are relatively easy to acquire (for neurofeedback purposes often with as few as 1-16 electrodes), the setup is noninvasive and unobtrusive, and consumer-grade systems for home use are already available (e.g., the Emotiv system).

The overall objective of the proposed project was to start exploring the potential of EEG-based neurofeedback as a novel treatment approach for stuttering. Given that the literature to date contains no publications of any prior efforts toward this goal, our project involved primarily early stage, exploratory work that sought to determine the general

feasibility of this approach. Nevertheless, to the extent possible, these initial efforts were strongly grounded in the current knowledge of neurobiological deficits underlying stuttering as well as the extant literature on neurofeedback treatments for other neurodevelopmental disorders and neurological conditions. Based on this foundation, more specific objectives of the proposed project were to investigate (a) whether adults who stutter can learn to voluntarily regulate aspects of brain activation throughout a series of neurofeedback training sessions, and (b) whether the regulation of brain activity targeted here has any measurable effects on the frequency (in percent stuttered syllables) of stuttering moments.

In order to discuss the rationale and supporting evidence for these objectives in more detail, it is necessary to point out a few important highlights from the general literature on clinical applications of EEG neurofeedback. Across those studies, two types of brain signals have been widely targeted for self-regulation: slow cortical potentials (SCP) and the sensorimotor rhythm (SMR). SCPs, the signals also targeted in the present work, reflect slow changes (from 300 ms to multiple seconds) in the EEG signal. Based on theoretical models and indirect empirical evidence, other authors have proposed that, from a functional perspective, SCPs may reflect a regulation mechanism for the excitation thresholds of pyramidal cells in the cortex, with increasing negativity corresponding to increased excitability and increasing positivity corresponding to decreased excitability (Birbaumer, 1999; Elbert, 1990).

Regardless of whether or not this hypothesis regarding the relationship between scalp-recorded SCPs and underlying neurophysiology is correct, SCP self-regulation has proved to be an effective treatment method for, among other conditions, ADHD and

epilepsy. For example, numerous studies on children with ADHD confirmed a reduction in symptoms after EEG-based neurofeedback training (e.g., Heinrich et al., 2004; Liechti et al., 2012). During tests after completion of the training, symptoms were reduced even though the children were not instructed during these tests in which direction (i.e., toward negativity or positivity) to shift their SCPs. Similar self-regulation of SCPs has also been shown to be successful in the treatment of epilepsy: nearly two-thirds of patients have been able to gain control over their SCPs and reduce the occurrence of seizures, again without requiring specific instruction as to whether SCPs should change toward negativity vs. positivity (e.g., Kotchoubey et al., 2001; Rockstroh et al., 1993).

Lastly, it should be specified that the neurofeedback signal selected here for training (i.e., SCPs) was not selected based on any specific relationship to stuttering. To date, no specific electrophysiological correlates of stuttering are known (neither for stuttering as a disorder nor for individual stuttering moments). Our interpretation, based on the fact that altering SCPs has resulted in significant changes across many clinical populations (e.g., epilepsy, ADHD, Tourette Syndrome, etc.) for which the SCP signal was also not specifically related to the underlying mechanisms, is that the brain may be able to transfer its learned regulation of one aspect of brain activity to other processes and systems which underlie the symptoms of the disorders.

Given (a) the reported overactivation of cortical systems in stuttering (see Background and Rationale), (b) the above clinical results on ADHD and epilepsy, and (c) numerous suggestions of EEG similarities in ADHD, epilepsy, and stuttering and even reports of the co-occurrence of epilepsy and stuttering (e.g., Ratcliff-Baird, 2001; Sechi, Cocco, D'Onofrio, Deriu, & Rosati, 2006), neurofeedback training that facilitates the

ability to self-regulate SCPs may benefit individuals who stutter. Here, three adults who stutter trained with a neurofeedback paradigm based on the overall level of negativity vs. positivity of the SCPs. The primary aims of the study were to establish the feasibility of this approach, to examine potential outcome measures, and to explore the time course of improvement, if any, within a relatively short 6-week training period.

Method

Participants

Three participants, ranging from 19 to 42 years of age, with histories of persistent developmental stuttering were recruited from the Seattle area via the University of Washington Communication Studies Participant Pool and through general advertising. All participants gave informed consent, and the protocol was approved by the University of Washington Institutional Review Board.

Participants had received a variety of treatments in the past, but none were receiving treatment at the time of the study. All were fluent speakers of American English, and one was bilingual. No clinically significant additional speech, language, or hearing problems were reported.

Participant 1 (SM49) was a 42-year-old right-handed male who began stuttering at the age of 2 ½ years. He reported being a monolingual English speaker, with no history of speech, language, or hearing problems other than stuttering. He reported no history of neurological or psychological problems, and was not taking any medications. He was an active member of the National Stuttering Association (NSA), but had not recently participated in any stuttering treatment.

Participant 2 (SM56) was a 19-year-old right-handed male who began stuttering in early elementary school. He reported learning English and Japanese simultaneously, but described American English as his primary language. He recalled difficulty with articulation of /r/ in early elementary school, but reported no current hearing, speech, or language difficulties other than stuttering. His most recent stuttering treatment was more than a year prior to participation in the present study. He denied a history of neurological or psychological problems, and was taking medication only for allergies.

Participant 3 (SM51) was a 27-year-old male who began stuttering around the age of 4. He described himself as right-handed, but commented that he used his left hand to write. He was a monolingual English speaker with no history of speech, language, or hearing problems other than stuttering. He also reported no history of neurological or psychological problems, and was not taking any medications.

For each participant, samples of conversational speech and reading were audio- and video-recorded during an initial screening for offline stuttering assessment by means of the Stuttering Severity Instrument – 4th Edition (SSI-4; Riley 2009). The SSI-4 provides an overall severity score that is based on subscores reflecting the percent of stuttered syllables during conversation/monologue and reading, the average duration of the three longest stuttering moments, and the severity of secondary characteristics (e.g., lip pressing, eye blinking, head movements, etc.).

During administration of the SSI-4, SM49 stuttered on 12% of syllables in conversation/monologue (36/300) and 1.44% of syllables in reading (4/278). The average duration of his longest three stuttering moments was 2 seconds, and his secondary characteristics were rated 8. He was scored “not noticeable unless looking for it” for

Distracting Sounds, “severe and painful looking” for Facial Grimaces, “barely noticeable to casual observer” for Head Movements, and “none” for Movements of the Extremities. The overall SSI-4 score for SM49 was 28, which corresponds to a percentile rank of 61-77 and an overall severity rating of “Moderate.”

SM56 stuttered on 5.67% of syllables in conversation/monologue and 10% of syllables in reading. The average duration of his longest three stuttering moments was 1 second, and his secondary characteristics were rated 3. He scored only for Head Movements which were “distracting.” SM56’s overall SSI-4 score was 21, corresponding to the percentile range 24-40 and an overall severity rating of “Mild.”

SM51 stuttered on 12.67% of syllables in conversation/monologue and 4.4% of syllables in reading. The average duration of his longest three stuttering moments was 2 seconds, and his secondary characteristics were rated 11. He was scored “very distracting” for Distracting Sounds, “barely noticeable to casual observer” for Facial Grimaces and Head Movements, and “distracting” for Movements of the Extremities. Taken together, these subscores yielded an overall SSI-4 score of 32, a percentile rank of 78-88, and an overall severity rating of “Severe.”

Procedure

Participants were unaware of any potential treatment aspects of the study, and they were also not familiarized with general information about any clinical applications of neurofeedback until after the study. Each participant completed twelve 1-hour sessions of neurofeedback training distributed as evenly as possible over a 6-week period (depending on the subject’s availability and schedule). This number of training sessions was deemed

to be a reasonable compromise between the amount of training used in previously published work and practical limitations associated with an initial pilot study (including potential difficulties with subject recruitment for a longer period of time). No sham feedback conditions were used in this initial pilot study.

Instrumentation

During each training visit to the lab, subjects sat inside a sound booth in front of a computer monitor while wearing a 32-channel electrode cap (Neuroscan Quik-Cap). A small subset of the electrodes were used, namely the vertex electrode Cz, a reference electrode at the tip of the nose, and a ground electrode high on the forehead. Horizontal and vertical electrooculography (EOG) electrodes were used to reject trials contaminated by horizontal or vertical eye movements. The electrodes were connected to an integrated system for filtering and amplifying electrophysiological data (Grass Model 15 with 15A54 amplifiers). The data were filtered with band-pass cut-off frequencies of 0.01 and 30 Hz before being sampled at a rate of 256 samples per second. Standard skin preparation procedures were used to maintain electrode impedances below 5k Ω as verified with an electrode impedance meter (Grass F-EZM4). Data acquisition, data processing, and the real-time neurofeedback display were controlled by custom MATLAB code.

In each training session, participants completed at least three blocks of 40 trials. Each trial was 9 seconds in duration (2 seconds baseline, 7 seconds active). On the computer monitor, participants viewed a scene with a hot air balloon. At the beginning of each trial, an arrow (pointing either up or down) appeared for two seconds, and baseline SCP and eye data were collected. Participants were instructed to stare at the center of the screen

and to avoid blinking and eye movements during this baseline phase. The arrow then disappeared, and the position of the balloon started varying with the participant's SCP. Participants were verbally instructed to try and make the balloon move in the direction indicated by the arrow.

To provide online neurofeedback, the voltage of the filtered signal from the Cz electrode was averaged or "smoothed" over 64 data samples (250 ms) in the time domain, and the vertical position of the balloon was updated every 32 data samples (125 ms). In other words, the screen was updated based on a running average with overlapping windows. Increasing SCP negativity relative to baseline caused the balloon to move up whereas increasing positivity relative to baseline caused the balloon to move down.

At the end of each trial, a score on the screen indicated whether or not that trial was successful (i.e., whether SCP voltage had sufficiently increased or decreased, averaged across the entire trial, to meet the pre-determined criterion). The criterion for success depended on the level of the game: with increasing levels of the game, increasingly larger changes in the SCP were required for the trial to be considered successful. For example, at Level 1, the criterion for gaining a point was 2 μV and the criterion for losing a point was -2 μV . At Level 2, the criteria were 2.2 and -2.2 μV , respectively. Participants gained one point for a successful trial and lost a point for an unsuccessful trial; neutral trials (i.e., those with no change) resulted in no change to the score. After 10 successful trials, participants advanced to the next higher level. If at least 8 trials were non-successful within a block of 10 trials, the participant returned to the previous level. Within a given level, scores could reach negative values, but the lowest possible level was Level 1.

Trials during which participants blinked or made eye movements in the baseline phase were excluded and reattempted during an additional block of trials. The feedback display system was programmed such that blinking and eye movements were counterproductive to a participant's score. To minimize the possibility of participants using such movements to influence the EEG data, the program implemented a correction algorithm that subtracted from the EEG voltage, on a sample-by-sample basis, a value equivalent to 10% of the EOG voltage.

Measures

Subjects were asked, at the beginning of each session, to rate their stuttering severity over the last 3-4 days on a scale of 1 to 10, with 1 being very fluent (i.e., without any stuttering), and 10 being very dysfluent (i.e., the most severe stuttering they had experienced). Immediately before and after each training session, samples of conversational speech and reading were audio- and video-recorded to calculate the percentage of syllables stuttered (%SS). One of six reading passages, consistent in readability (including number of syllables, words, and sentences; Flesch, 1951) as well as Brown's word weights (Brown, 1945), was presented on the computer monitor. The sequence of six passages was used four times, for a total of 24 passages (i.e., pre- and post-readings in each of 12 sessions), with the order of the six passages within the sequence randomly varying across the participants. A set of conversation topics had been established for use with all participants. For each sample, the participant was asked to speak about the topic for approximately two minutes (the experimenter asked follow-up questions, however, and therefore these samples are referred to as conversational samples rather than monologues). Most samples reached 300 syllables in length, but some

samples were shorter. An undergraduate research assistant transferred the video files to a computer and assigned codes that allowed blind analyses of stuttering frequency. Due to problems with this transferring and coding process, 24 of the 72 samples (3 subjects \times 12 sessions \times 2 recordings [pre and post]) were unavailable for analysis.

The present author analyzed all reading and conversational speech samples while remaining blind to the order of the samples within the 6-week training program and the pre-training vs. post-training status of all samples. The author's inter-rater reliability (relative to the laboratory's Principal Investigator) for syllable-by-syllable stuttering frequency counts as determined by the Kappa index (Cohen, 1960) had been previously determined to be .91. For the present purposes, the following types of speech behaviors were counted as stuttering moments: monosyllabic word repetitions (e.g., "**And and and and** then we went to a baseball game"), audible or inaudible sound prolongations (e.g., "I have ssssssstuttered since I can remember" or "I -----took a course in physics"), sound or syllable repetitions (e.g., "I lived on a **f-f-f-farm**" or "I went to **el-el-el-elementary** school in Seattle"), and broken words (e.g., "I was in the fifth **gra—ade**").

Given the exploratory nature of this pilot work and the small number of subjects, we examined training-related changes in stuttering frequency by means of single-case analyses rather than inferential statistics.

Results

PARTICIPANT 1 (SM49)

Game Data

SM49 was unable to progress through different levels of the neurofeedback game. At the end of the 12 sessions, he remained at Level 1, with a score of -7.

EEG Data

SM49's EEG data demonstrate that he did not learn to regulate SCPs either within (different shades of blue and red) or across (different panels) the 12 training sessions (Figure 1).

Speech Data

Self-Rating

SM49 reported relatively consistent global self-ratings of stuttering severity across the 6-week period. With the exception of the 10th visit to the lab—during which he reported a self-rating of 4--SM49 always self-rated his severity as 3 on our scale from 1 to 10. Thus, SM49 perceived no change in his stuttering as a result of the training sessions (Figure 2).

Conversational samples across training sessions

Across conversational samples, SM49's stuttering frequency showed considerable variation (Figure 3, top panels). During pre-training samples, stuttering frequency ranged from 6% SS (training session 11) to 22% SS (training session 7). On average, he stuttered on 11.5% of syllables before training, with a standard deviation of 4.55%. Across post-training conversational samples, SM49's stuttering frequency ranged from 6.67% SS (training session 2) to 17.67% SS (training session 3). On average, he stuttered on 11.39% of syllables after training, with a standard deviation of 3.34%. As graphically

summarized in Figure 3, SM49's stuttering frequency during conversation was highly similar for the post-training and pre-training recordings.

With regard to potential data trends over time, SM49's conversational samples failed to show a relationship between stuttering frequency and the amount of neurofeedback training for both the post-training ($R^2 = 0.02$) and pre-training ($R^2 = 0.07$) recordings.

Reading samples across training sessions

SM49 stuttered considerably less during reading (Figure 3, bottom panels) as compared with conversation. In reading samples recorded prior to each training session, SM49's stuttering rates ranged from 0.33% SS (training session 5) to 5% SS (training session 1). An average of 2.05% of syllables was stuttered in reading samples recorded pre-training, with a standard deviation of 1.26%. In reading samples recorded post-training, stuttering rates ranged from 1% SS (training session 4) to 9.33% SS (training session 9). On average, he stuttered on 3.11% of syllables post-training, with a standard deviation of 2.22%. Thus, SM49's stuttering frequency was higher in post-training samples, but it is clear from Figure 3 that this increase was largely due to an outlier data point in session 9.

Examining potential changes in stuttering frequency data across all 12 training sessions, the trend lines and associated R^2 values in Figure 3 again suggest that, for SM49, neurofeedback training was not associated with changes in stuttering frequency. Measures of percent stuttered syllables did not vary with the amount of training for either post-training samples ($R^2 = 0.04$) or pre-training samples ($R^2 = 0.00$).

PARTICIPANT 2 (SM56)

Game Success

SM56 was highly successful in terms of progressing through the various levels of the neurofeedback game. Although he experienced limited success during the early training sessions, proceeding slowly through Levels 1 and 2, his score started to show faster improvement from approximately the 8th training session. At the end of the 12 sessions, SM56 had reached Level 18.

EEG Data

The first three panels of brain data in Figure 4 indicate that, initially, SM56 showed no considerable changes in brain activity based on the provided visual feedback. However, he did demonstrate learning in the 4th and 5th sessions. The corresponding panels of Figure 4 show an increased separation between the red lines (trials in which the goal was a more negative SCP) and blue lines (trials in which the goal was a more positive SCP). Although this separation mostly disappeared again for training sessions 6 and 7, it then re-appeared and was maintained throughout all subsequent training sessions. It should be noted, however, that sessions 8-12 were associated with voltage changes that were more both more extensive and more rapid than one would expect based on typical EEG data. From the available data, it cannot be determined with certainty to what extent these SCP changes are due to ocular, muscle, or other artifacts. For example, panels 9 (for trials aiming for positivity, shown in blue) and 12 (for trials aiming for negativity, shown in red) suggest that the large changes in SCP data (already adjusted

based on the EOG signal, see Method) are not always associated with similar changes in the EOG signal.

Speech Data

Self-Rating

As shown in Figure 5, SM56 reported self-ratings of stuttering severity ranging from 2 to 4 (i.e., with 1 least severe and 10 most severe). He reported self-ratings of 2 twice, during training sessions 2 and 11, and self-ratings of 4 twice, during sessions 3 and 7. All other self-ratings were a 3. Slightly more fluctuation was observed during the first half than the second half of the 6-week period.

Conversational samples across training sessions

SM56's stuttering frequency in the conversational samples (Figure 6, upper panels) ranged from 4% SS (training session 8) to 9.67% SS (training session 12) in pre-training measurements. On average, he stuttered on 6.96% SS pre-training, with a standard deviation of 1.99%. Post-training data were very similar to pre-training data, both in terms of the range and average. In post-training samples, SM56's stuttering frequency ranged from 3.67% SS (training session 8) and 9.3% SS (training sessions 11 and 12). He averaged 7.20% of syllables stuttered post-training, with a standard deviation of 2.21%. Figure 6 illustrates that SM56 had generally unchanged stuttering frequency rates for conversation post-training as compared with pre-training. Over the course of the 12 training sessions, SM56's stuttering frequency also remained essentially unchanged, and this was true for both the pre-training ($R^2 = 0.01$) and post-training ($R^2 = 0.05$) speech

samples. The low R^2 values clearly indicate that stuttering frequency did not vary with the amount of neurofeedback training.

Reading samples across training sessions

Unlike the situation for his conversational data, SM56's stuttering frequency during reading did decrease in post-training samples as compared with pre-training samples (Figure 6, bottom panels). In pre-training reading samples, SM56 demonstrated a range of stuttering frequencies from 3.33% SS (training session 5) to 9.67% SS (training sessions 5 and 10). His average stuttering frequency was 6.99% SS pre-training, with a standard deviation of 2.19%. Post-training, this stuttering frequency was slightly reduced, ranging from 3% SS (training session 8) to 7% SS (training session 2), with an average of 5.2% SS and a standard deviation of 1.22%. When considering data trends over time, on the other hand, there was no change in the reading-based percent stuttered syllables for either post-training ($R^2 = 0.00$) or pre-training ($R^2 = 0.00$) samples.

PARTICIPANT 3 (SM51)

Game Data

SM51's success with the program, in terms of game score, was gradual but consistent. He completed his final training at Level 5 with a score of 1.

EEG Data

As analyzed here (averaged across sets of 20 trials), the EEG data for SM51 show no clear evidence of learning either within training sessions (represented by different shades

of blue and red) or across the 12 sessions (represented by different panels). In other words, this participant's SCP data did not show a noticeable separation between trials aimed toward positivity vs. negativity (Figure 7).

Speech Data

Self-Rating

SM51 reported global self-ratings of stuttering severity ranging from 4 to 6 (Figure 8). He reported a self-rating of 4 once, during training session 2, and self-ratings of 6 three times, during sessions 1, 8 and 9. All other self-ratings were a 5.

Conversational samples across training sessions

SM51's stuttering frequency data for all conversational samples are shown in the upper panels of Figure 9. He stuttered on 8.62% of syllables on average (standard deviation 3.20) in the pre-training conversational samples where stuttering frequency ranged from 4.3% SS (training session 9) to 14.3% SS (training session 4). Across the post-training conversational samples, his average stuttering frequency was 9.41% SS (standard deviation 2.29%) and the range was from 6.67% SS (training session 9) to 15% SS (training session 11). Thus, SM51 showed similar stuttering frequency measures for post-training samples as compared with pre-training samples.

Analyzing data trends over time, SM51's stuttering frequency showed only a very weak relationship with the amount of training for the post-training speech samples ($R^2 = 0.18$) and even less of an indication of a relationship for the pre-training speech samples ($R^2 = 0.09$).

Reading samples across training sessions

SM51 stuttered only very mildly when reading aloud, as shown in the bottom panels of Figure 9. On average, he stuttered on 1.38% SS pre-training, with a standard deviation of 0.71% and a range from 0.67–2.67% SS. Stuttering frequency for the post-training reading samples ranged from 0.67% SS (training session 7) to 3% SS (training session 1), with an average of 1.66% SS and a standard deviation of 0.8%. Thus, overall, stuttering frequency remained similar in post-training reading samples as compared with pre-training reading samples, but it should be noted that this participant exhibited only minimal stuttering during such reading samples.

Across the 12 sessions, SM51's stuttering frequency data from the post-training reading samples showed a trend to decrease in stuttering frequency ($R^2 = 0.29$) whereas the pre-training reading samples showed essentially no relationship with the amount of training ($R^2 = 0.09$).

Discussion

The purpose of the present study was to collect pilot data regarding the feasibility of using neurofeedback as a potential technique in the treatment of individuals who stutter. The methods and procedures were based on principles and training protocols that have been documented in past research studies with a variety of clinical populations, including neurodevelopmental disorders and neurological conditions.

Three adults who stutter participated in a 12-session neurofeedback training program that aimed to gain voluntary control over the positivity vs. negativity of SCPs. Besides

the EEG data, we also collected behavioral data regarding (a) the achieved performance level in the computer game that served as the visual feedback channel, (b) self-ratings of stuttering severity, and (c) stuttering frequency measures during conversation and oral reading. Prior to the training sessions, participants were not informed that neurofeedback has been used as a treatment technique for any clinical populations (this information was provided only after the 12 sessions had been completed).

We interpret the obtained data as indicating that at least one participant (SM56) demonstrated some SCP learning during the program. This participant showed both a progression through several difficulty levels of the training game (eventually reaching Level 18) and the ability to regulate SCPs. The latter interpretation is offered with caution, however, given that the rate and extent of SCP voltage changes during some of the training sessions leave open the possibility that the SCP signals were contaminated by EOG or other artifacts. For this “successful” SCP learner, stuttering frequency during oral reading (but not conversation) was reduced in post-training recordings vs. pre-training recordings (recall that pre- and post-training refers to measurements made within, rather than across, individual training sessions). Thus, at least for reading, successful SCP learning may have contributed to short-term gains in speech fluency. In terms of long-term gains, however, these limited data from 12 training sessions did not provide any indication of a trend toward reduced stuttering rates across the sessions.

Another participant, SM51, was moderately successful in advancing through the difficulty levels of the training game (reaching Level 5), although this progress was not clearly observable in the actual SCP signals as graphically represented here (i.e., averaged across sets of 20 trials per session). This participant stuttered very little (< 2%

SS) during oral reading, and—unlike SM56 who had substantially higher stuttering rates during the same speech task—SM51 did not show short-term training benefits in the form of reduced stuttering rates in post-training vs. pre-training recordings. Interestingly, however, this participant's post-training reading samples showed the strongest observed, albeit still relatively weak, relationship between stuttering frequency and the amount of neurofeedback training ($R^2 = .29$).

Lastly, participant SM49 was not successful at advancing beyond Level 1 of the training game, and he also showed no evidence of SCP learning. This lack of learning was associated with (a) an absence of differences in stuttering frequency between pre- and post-training recordings (a small increase for the reading samples was attributable to a single outlier data point) as well as (b) an absence of a relationship between amount of training and stuttering frequency during either conversation or reading.

In summary, findings from the current study provide some, albeit very limited, support for further exploration of neurofeedback training as a potentially useful technique in the treatment of stuttering. Two of three subjects showed some degree of behavioral and/or SCP evidence of neurofeedback-based learning, and these same two subjects—but not the remaining third subject—showed at least limited evidence of changes in measures of stuttering frequency during oral reading. For one of these two subjects (SM56), reading-based stuttering frequency was lower in recordings made immediately after the training sessions as compared with recordings made immediately before the training sessions. For the other subject (SM51), reading-based stuttering frequency showed a trend to decrease across the total series of 12 training sessions. Given these results for oral reading, it might seem surprising—or even problematic—that stuttering frequency

during conversational speech did not show any such evidence of pre- to post-training changes or trends to decrease across repeated training sessions. It should be noted, however, that earlier or larger improvements in stuttering for reading vs. conversation are also commonly observed for other treatment approaches. For example, it is well documented that the fluency-enhancing effects of altered auditory feedback are substantially larger when the speaking task involves oral reading as opposed to conversation or monologue (Armson & Stuart, 1998; Foundas, Mock, Corey, Golob, & Conture, 2013).

In addition to such variability inherent in stuttering, a number of methodological limitations may have affected the outcome of the present pilot study. First, we have explored here only one type of neurofeedback training; namely, SCP training. It is of course possible that training protocols focusing on other aspects of brain activity may be more effective. Currently ongoing work in our laboratory is already exploring protocols based on frequency-band analyses (in particular sensorimotor rhythm) and hemispheric lateralization. Second, for practical reasons (e.g., participants' availability and willingness to make repeated visits to the laboratory), our training protocol was limited to only 12 sessions distributed over 6 weeks. A larger number of training sessions, or a more intensive training with the same number of sessions distributed over a shorter period of time, may also be more effective. Third, to examine the effect of neurofeedback training on speech fluency, we focused here only on self-ratings of stuttering severity and measures of stuttering frequency. Additional measures such as, for example, the duration of individual stuttering moments might reveal more subtle changes as a result of the training. Fourth, given that the three subjects included here were the first three

individuals to complete neurofeedback training protocols in our laboratory, technical aspects of the feedback display system (e.g., data smoothing, image update rates, method for indicating targeted negativity vs. positivity) continued to be fine-tuned during the initial training sessions. Such adjustments, although minor, may have had subtle effects on participants' learning. Given these limitations, and the obtained preliminary results, it can be concluded that further studies regarding the feasibility of neurofeedback training for individuals who stutter are warranted.

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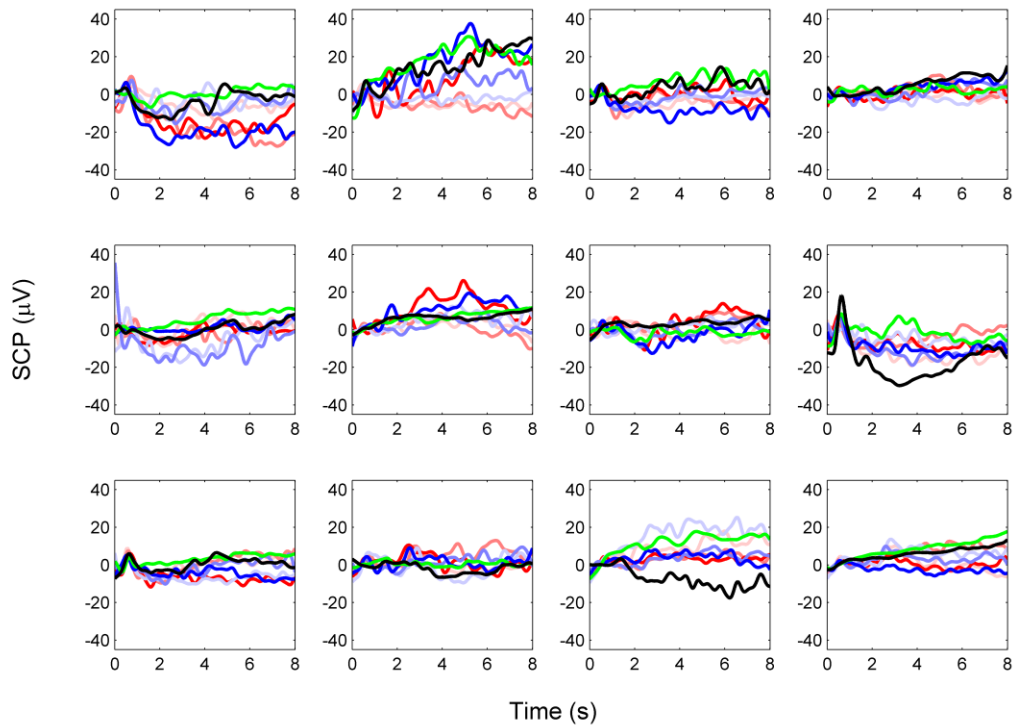


Figure 1. Participant SM49's slow cortical potentials for 12 sessions of neurofeedback training (sessions ordered from top left to bottom right by row). Red and blue lines represent EEG data averaged across each of 3 sets of 20 trials with negativity or positivity goals, respectively. The lightest, middle, and darkest shades of color mark the 1st, 2nd, and 3rd set of trials. Green and black lines represent averaged electrooculography data from the trial set with the largest deviation from baseline in the red or blue EEG data, respectively.

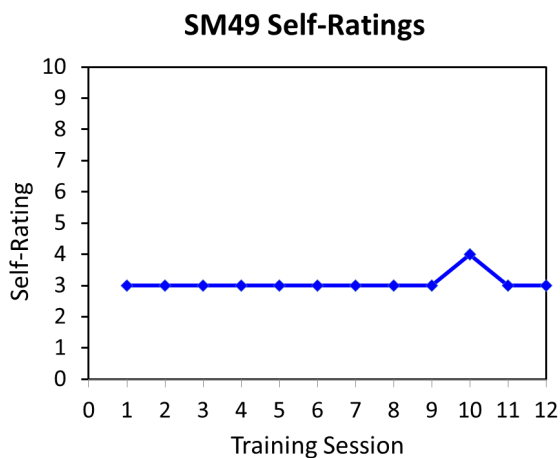


Figure 2. SM49's self-ratings of stuttering severity prior to each of 12 sessions of neurofeedback training. Stuttering severity was self-rated on a scale from 1 (no stuttering) to 10 (most severe stuttering ever experienced).

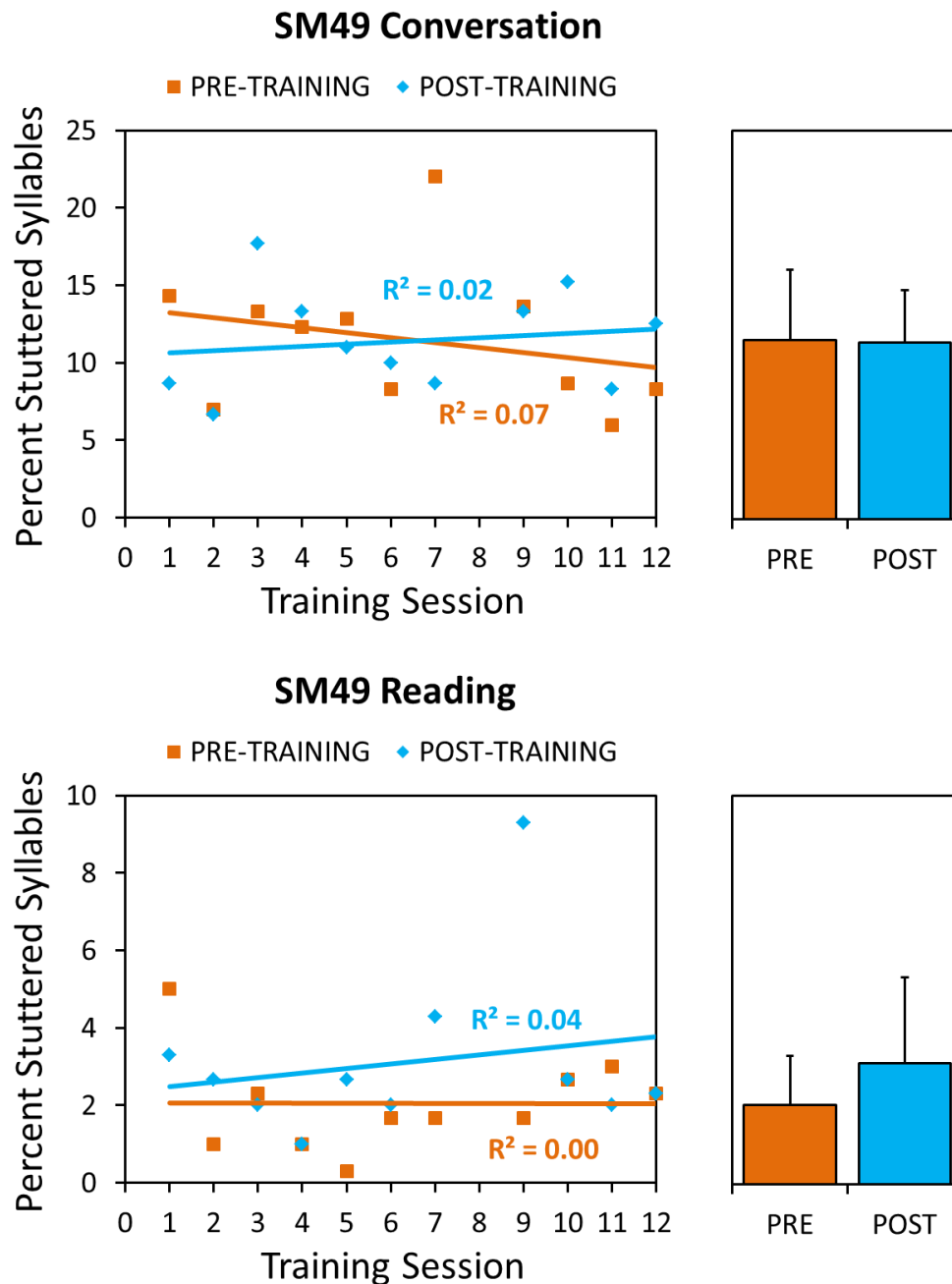


Figure 3. SM49's stuttering frequency (in percent stuttered syllables, %SS) during conversation (top) and oral reading (bottom) pre- and post-training for each of 12 neurofeedback sessions. Scatterplots indicate %SS data for each individual speech sample. Bar graphs indicate %SS means and standard deviation for conversational and reading samples averaged across all sessions.

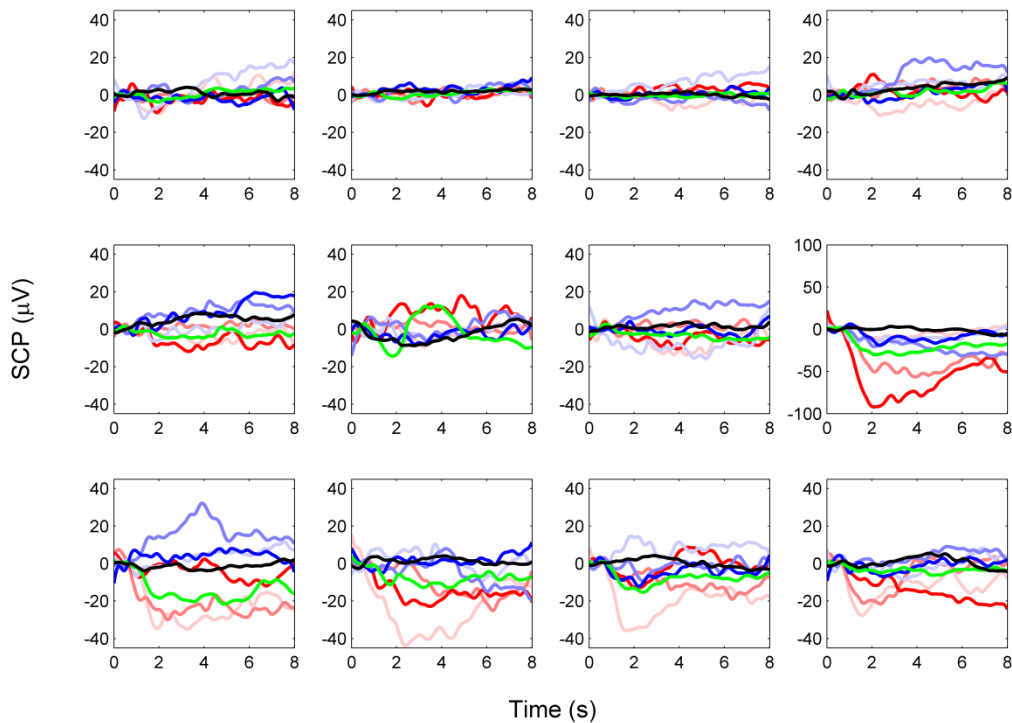


Figure 4. Participant SM56's slow cortical potentials for 12 sessions of neurofeedback training (sessions ordered from top left to bottom right by row). Red and blue lines represent EEG data averaged across each of 3 sets of 20 trials with negativity or positivity goals, respectively. The lightest, middle, and darkest shades of color mark the 1st, 2nd, and 3rd set of trials. Green and black lines represent averaged electrooculography data from the trial set with the largest deviation from baseline in the red or blue EEG data, respectively.

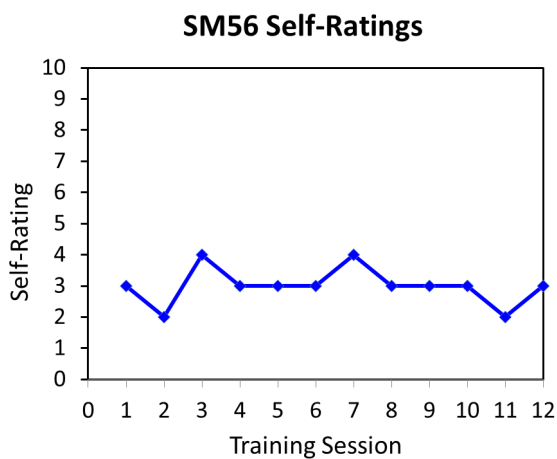


Figure 5. SM56's self-ratings of stuttering severity prior to each of 12 sessions of neurofeedback training. Stuttering severity was self-rated on a scale from 1 (no stuttering) to 10 (most severe stuttering ever experienced).

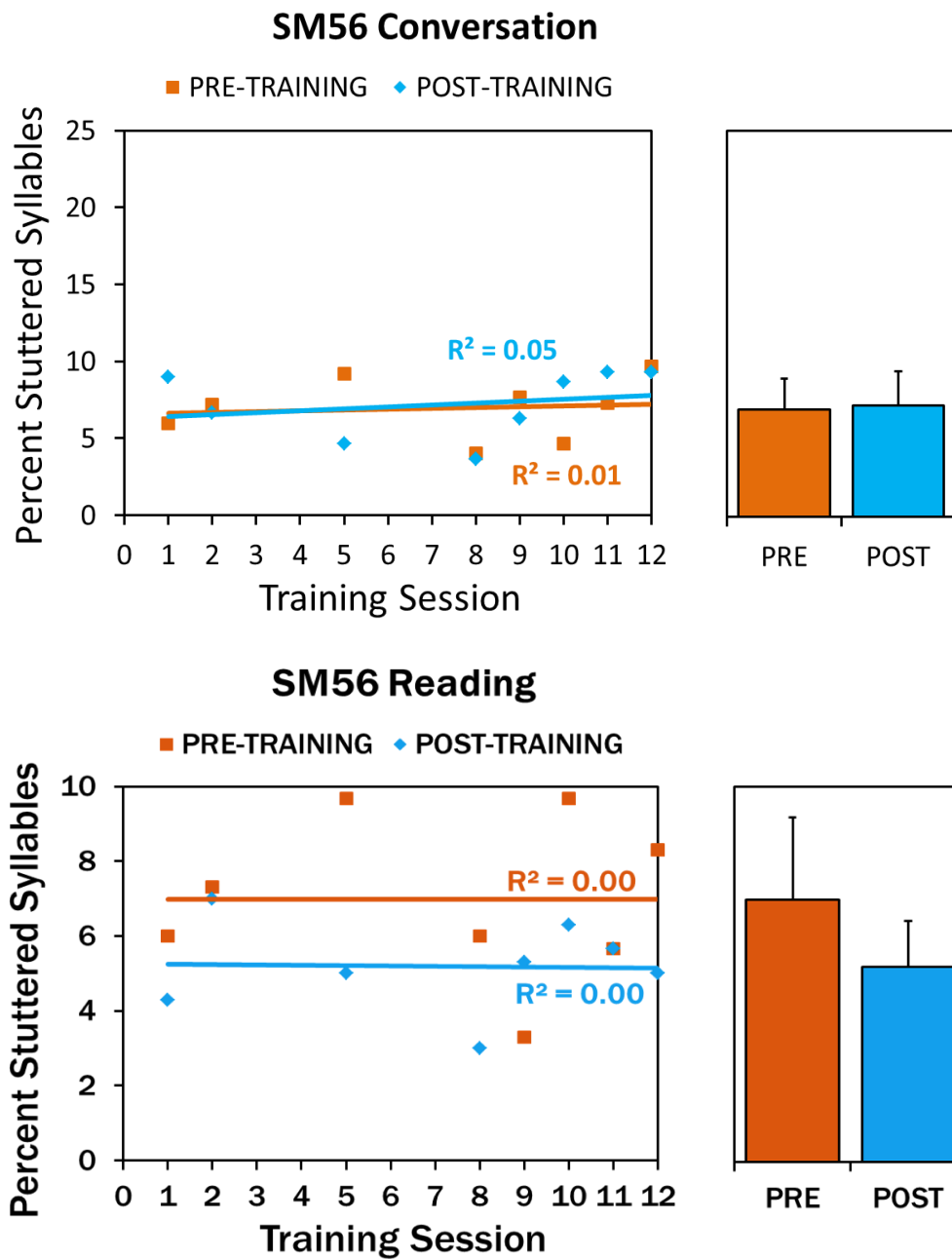


Figure 6. SM56's stuttering frequency (in percent stuttered syllables, %SS) during conversation (top) and oral reading (bottom) pre- and post-training for each of 12 neurofeedback sessions. Scatterplots indicate %SS data for each individual speech sample. Bar graphs indicate %SS means and standard deviation for conversational and reading samples averaged across all sessions.

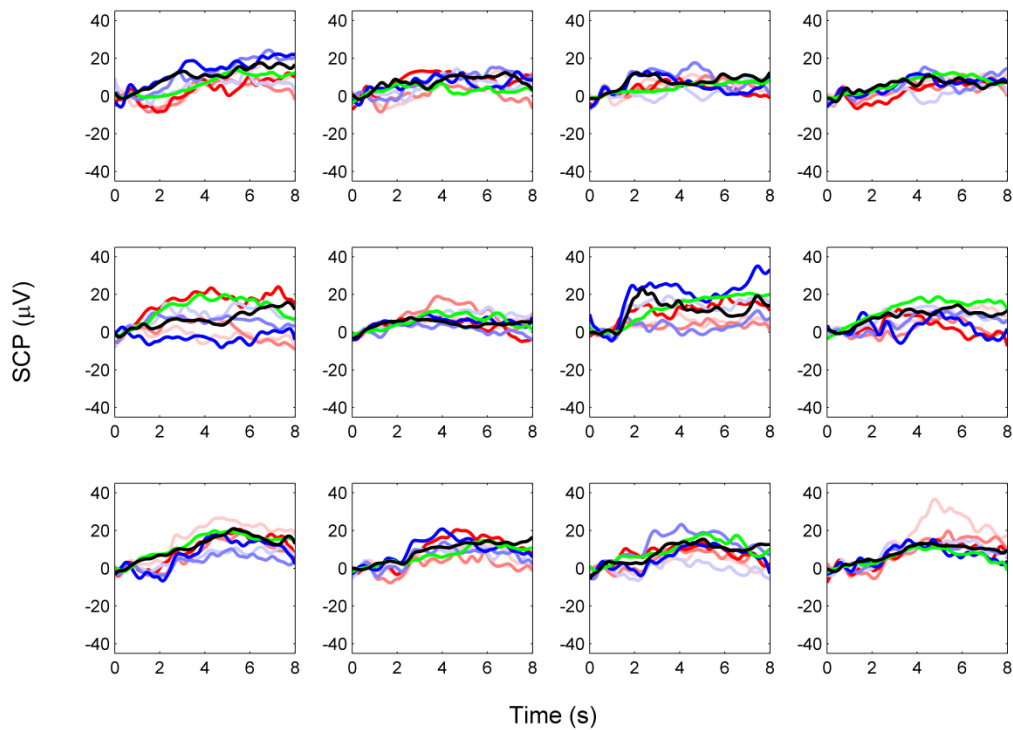


Figure 7. Participant SM51's slow cortical potentials for 12 sessions of neurofeedback training (sessions ordered from top left to bottom right by row). Red and blue lines represent EEG data averaged across each of 3 sets of 20 trials with negativity or positivity goals, respectively. The lightest, middle, and darkest shades of color mark the 1st, 2nd, and 3rd set of trials. Green and black lines represent averaged electrooculography data from the trial set with the largest deviation from baseline in the red or blue EEG data, respectively.

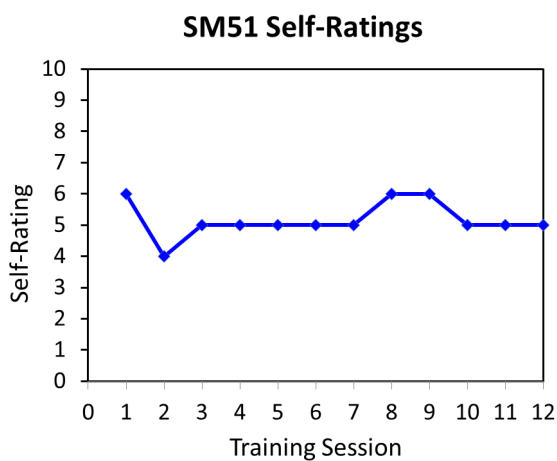


Figure 8. SM51's self-ratings of stuttering severity prior to each of 12 sessions of neurofeedback training. Stuttering severity was self-rated on a scale from 1 (no stuttering) to 10 (most severe stuttering ever experienced).

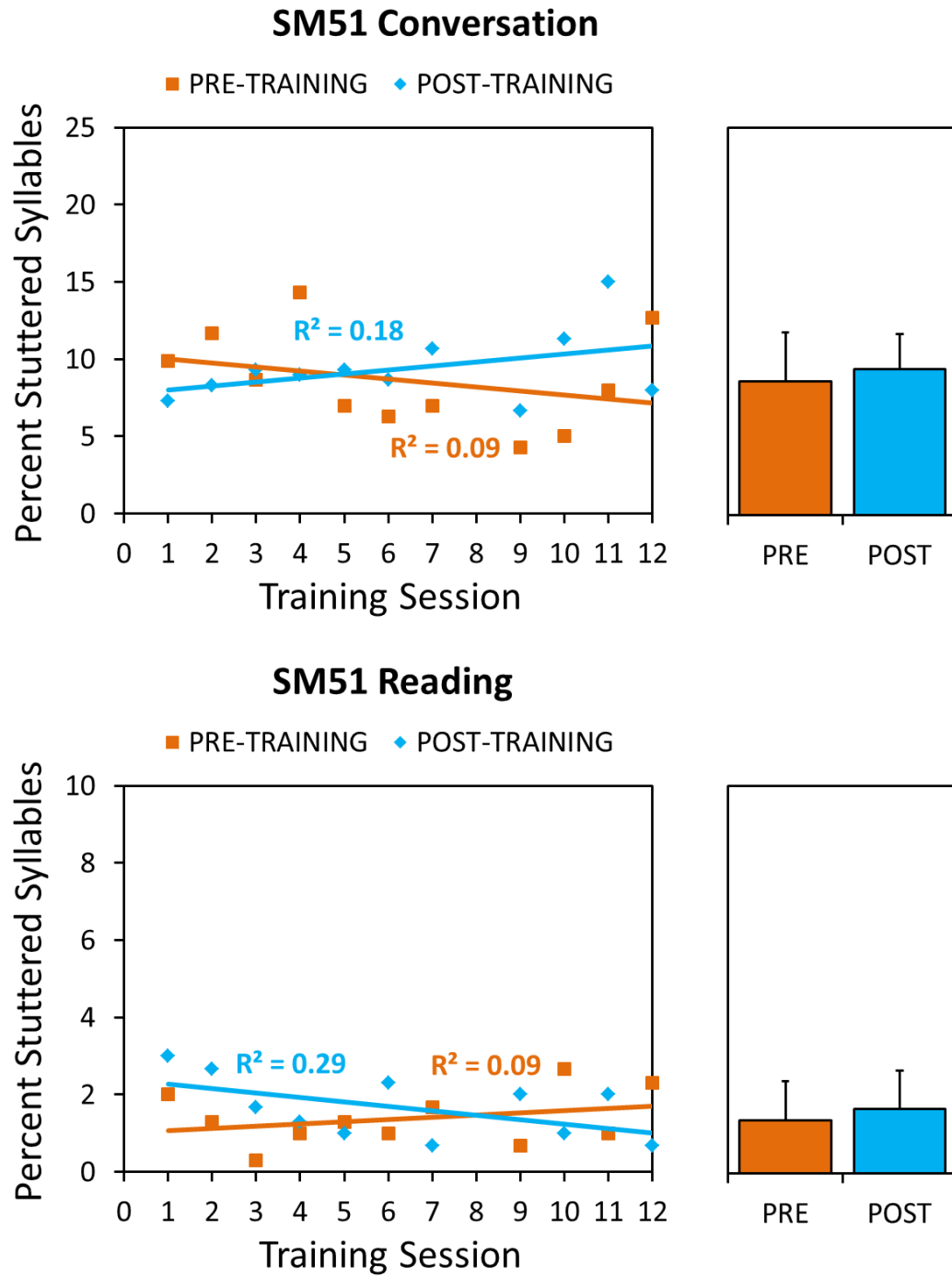


Figure 9. SM51's stuttering frequency (in percent stuttered syllables, %SS) during conversation (top) and oral reading (bottom) pre- and post-training for each of 12 neurofeedback sessions. Scatterplots indicate %SS data for each individual speech sample. Bar graphs indicate %SS means and standard deviation for conversational and reading samples averaged across all sessions.