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Direction of attentional focus in biofeedback treatment for /r/ misarticulation

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Structured Abstract

Background—Maintaining an external direction of focus during practice is reported to facilitate acquisition of nonspeech motor skills, but it is not known whether these findings also apply to treatment for speech errors. This question has particular relevance for treatment incorporating visual biofeedback, where clinician cueing can direct the learner's attention either internally (i.e., to the movements of the articulators) or externally (i.e., to the visual biofeedback display).

Aims—This study addressed two objectives. First, it aimed to use single-subject experimental methods collect additional evidence regarding the efficacy of visual-acoustic biofeedback treatment for children with /r/ misarticulation. Second, the study compared the efficacy of this biofeedback intervention under two cueing conditions. In the external focus (EF) condition, participants' attention was directed exclusively to the external biofeedback display. In the internal focus (IF) condition, participants viewed a biofeedback display, but they also received articulatory cues encouraging an internal direction of attentional focus.

Methods & Procedures—Nine school-aged children were pseudorandomly assigned to receive either internal or external focus cues during eight weeks of visual-acoustic biofeedback intervention. Accuracy in /r/ production at the word level was probed in three to five pre-treatment baseline sessions and three post-treatment maintenance sessions. Outcomes were assessed using visual inspection and calculation of effect sizes for individual treatment trajectories. In addition, a mixed logistic model was used to examine across-subjects effects including phase (pre/post-treatment), /r/ variant (treated/untreated), and focus cue condition (internal/external).

Outcomes & Results—Six out of nine participants showed sustained improvement on at least one treated /r/ variant; these six participants were evenly divided across EF and IF treatment groups. Regression results indicated that /r/ productions were significantly more likely to be rated accurate post-treatment than pre-treatment. Internal versus external direction of focus cues was not a significant predictor of accuracy, nor did it interact significantly with other predictors.

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Conclusions—The present results are consistent with previous literature reporting that visual-acoustic biofeedback can produce measurable treatment gains in children who have not responded to previous intervention. These findings are also in keeping with previous research suggesting that biofeedback may be sufficient to establish an external attentional focus, independent of verbal cues provided. The finding that explicit articulator placement cues were not necessary for progress in treatment has implications for intervention practices for speech sound disorders in children.

Introduction

Speech sound disorder, in which developmentally inappropriate errors affect the intelligibility and perceived typicality of spoken communication, has an estimated prevalence of approximately 4% among children in the early school-age years (Shriberg, Tomblin, and McSweeny, 1999). Most of these children will go on to achieve typical speech, spontaneously or through intervention. However, among children with a history of speech sound disorder, an estimated 30% continue to exhibit residual errors affecting sibilant and rhotic sounds at nine years of age, and roughly 9% show these errors from 12–18 years and beyond (Lewis and Shriberg, 1994). Clinicians have called for more effective interventions for such errors, especially misarticulation of the phoneme /r/, characterized as the most common residual error in English (Ruscello, 1995; Shriberg, 2010). Recent evidence suggests that speech sound errors that do not respond to conventional methods may be eliminated through *visual biofeedback treatment*.

Visual biofeedback treatment and residual /r/ errors

In visual biofeedback, instrumentation is used to obtain and present precise measurements of some aspect of physiology or behavior. These measurements are displayed in real time to the learner, with the goal of providing new insight into aspects of behavior that are typically below the level of conscious control (Davis and Drichta, 1980; Volin, 1998). In biofeedback intervention for speech, the learner views a model representing correct production of the target sound; he/she is encouraged to compare his/her own output against the model and make adjustments to achieve a closer match. Various biofeedback technologies have been used to treat speech disorders in children and adults, including ultrasound imaging (e.g., Adler-Bock, Bernhardt, Gick, and Bacsfalvi, 2007; Preston, Brick, and Landi, 2013; McAllister Byun, Hitchcock, and Swartz, 2014; Preston et al., 2014), electromagnetic articulography (e.g., Katz, McNeil, and Garst, 2010), and electropalatography (e.g., Gibbon, Stewart, Hardcastle, and Crampin, 1999). The present study investigates *visual-acoustic biofeedback*, in which learners compare a dynamic image of the acoustic signal of their own speech to a static image representing the target sound. Certain sound contrasts are signaled by different configurations of the resonant frequencies of the vocal tract (formants). In visual-acoustic biofeedback, formant locations are represented in real time with spectrograms (e.g., Shuster, Ruscello, and Smith, 1992; Shuster, Ruscello, and Toth, 1995) or linear predictive coding (LPC) spectra (McAllister Byun and Hitchcock, 2012).

The results of previous studies suggest that visual-acoustic biofeedback can be an effective form of treatment for residual errors. Two case studies described the successful use of spectrographic biofeedback in three speakers with persistent /r/ errors (Shuster et al., 1992;

Shuster et al., 1995). McAllister Byun and Hitchcock (2012) examined the effects of visual-acoustic biofeedback treatment on /r/ misarticulation in 11 children aged six to 12 years. This quasi-experimental study consisted of a preliminary phase in which participants received a modified traditional form of articulatory intervention, followed by a phase of biofeedback treatment with a real-time LPC speech spectrum. Progress was measured with word-level generalization probes administered at the beginning, midpoint, and end of the study. No effect of intervention was observed after the traditional treatment phase, but /r/ words elicited after biofeedback treatment were found to be acoustically and perceptually more accurate than at previous time points.

Despite the encouraging nature of the findings reported above, previous studies have been limited by small sample size and a low overall degree of experimental control. For example, McAllister Byun and Hitchcock (2012) measured the effects of visual-acoustic biofeedback treatment only in the context of a preceding period of traditional treatment. This leaves open the possibility that the gains in /r/ production accuracy observed during the biofeedback period were actually a late-emerging response to traditional intervention. The present study investigated whether gains comparable to those reported in previous literature would be observed when biofeedback was provided as the only experimental intervention. Furthermore, in McAllister Byun and Hitchcock (2012), traditional articulator placement cues continued to be incorporated throughout the course of biofeedback intervention, and it is not known whether these cues were essential to produce the observed gains. This is a question of clinical significance, and it is also important in connection with the theoretical rationale for biofeedback that has been posited within the framework of principles of motor learning, discussed below.

Principles of motor learning in speech intervention

Children’s difficulty mastering English /r/ is generally assumed to be related to the complex articulatory configuration required to produce the sound. While most speech sounds are formed with a single major constriction or narrowing of the vocal tract, /r/ involves two major lingual constrictions—one in which the anterior tongue raises to approximate the palate, and one in which the tongue root retracts to narrow the pharyngeal space—as well as lip rounding (Adler-Bock et al., 2007; Alwan, Narayanan, and Haker, 1997). Children with residual errors on late-developing phonemes such as /r/ have been found to perform significantly below typical peers on oral-motor measures such as diadochokinetic rate (Preston and Edwards, 2009), as well as articulation rate in connected speech (Flipsen, 2003). These findings suggest that it is appropriate to investigate intervention for /r/ misarticulation within the context of motor skill learning.

Principles of motor learning refer to well-established observations that certain practice conditions facilitate the learning of motor skills, compared to other conditions. For example, a greater number of practice trials results in greater retention and generalization than a smaller number of trials (e.g., Shea and Kohl, 1991), and practicing different versions of a movement pattern (variable practice) produces greater learning than practicing only one version of the pattern (constant practice; e.g., Wulf and Schmidt, 1997). A large body of literature exists on the factors that enhance learning of nonspeech motor skills such golf swings and balance tasks (see Schmidt and Lee, 2005, for in-depth review). Recent years have seen increasing attention to the idea that principles of motor learning may also be relevant to intervention for speech disorders (e.g., Edeal and Gildersleeve-Neumann, 2011; Maas and Farinella, 2012; Maas, Gildersleeve-Neumann, Jakielski, and Stoeckel, 2014; see Maas et al., 2008, for review). The limited available evidence regarding such extension to the speech domain is promising (e.g., Edeal and Gildersleeve-Neumann, 2011; Knock, Ballard, Robin, and Schmidt, 2000). However, other findings indicate that not all children with motor-based speech disorders show enhanced learning in the conditions thought to be optimal for nonspeech motor learning (e.g., Maas, Butalla, and Farinella, 2012; Maas and Farinella, 2012). There is thus a need for direct study of different motor practice conditions in the speech domain, and in different populations.

Direction of attentional focus

Previous work has suggested that the efficacy of biofeedback treatment might be related to its ability to induce an external, as opposed to internal, direction of attentional focus (Shea and Wulf, 1999). *Internal focus* (IF) refers to a situation in which attention is directed to the movements and sensations of the body parts involved in the movement, such as the angle of the elbow and the position of the hands when holding a golf club. *External focus* (EF) is when attention is directed to more distal aspects, such as the weight and movement of the club (e.g., Wulf et al., 1998). In both cases, the movement *goal* is the same (e.g., to get the ball in the hole).

A range of studies have investigated the role of EF versus IF on performance in nonspeech motor learning tasks (e.g., Wulf, Höß, and Prinz, 1998; Wulf, McNevin, and Shea, 2001). Studies of both novice and expert learners performing a variety of nonspeech tasks have reported that EF can facilitate acquisition and retention of motor skills (see Wulf, 2007, for review). While most of these studies investigated movements involving extremities or the entire body, Freedman, Maas, Caligiuri, Wulf, and Robin (2007) found a similar benefit of EF in a nonspeech oral motor task involving application of a precise degree of force to a pressure bulb with the tongue. Furthermore, Lisman and Sadagopan (2013) compared unimpaired adult speakers' production of tongue twisters and novel utterances in an IF condition, in which participants were encouraged to focus on the feeling and movements of their articulators, versus an EF condition, in which participants were encouraged to focus on the sound of their speech. Participants showed greater variability and slower speech rates in the IF condition, suggesting that the advantage for EF may extend to motor learning in the speech domain. To date, it appears that no literature has directly investigated the hypothesis that manipulating direction of focus might influence speech-motor learning in children with speech sound disorder or residual speech errors. The present study investigated a manipulation affecting direction of attentional focus within the context of visual-acoustic biofeedback treatment for /r/ misarticulation.

Biofeedback intervention and direction of attentional focus

The advantage for EF over IF has been cited as part of the rationale for using visual biofeedback to treat speech disorders (e.g., McAllister Byun and Hitchcock, 2012). However, the methodology of previous studies investigating biofeedback has left ambiguity as to whether participants' focus was directed internally or externally. In the visual-acoustic biofeedback studies by Shuster et al. (1992) and McAllister Byun and Hitchcock (2012), the use of a real-time visual display encouraged an external direction of attentional focus, but participants also received explicit cues for articulator placement, which directed attention internally. Previous research has investigated whether instructions encouraging IF or EF can influence performance when the nature of visual feedback is held constant. Participants in the study of tongue movements by Freedman et al. (2007) all saw the same visual feedback display, but they received different instructions (e.g., "Focus on your tongue" for the IF group versus "Focus on the pressure bulb" for the EF group). In that study, significant group differences were observed in connection with the difference in focus instructions. Similarly, Shea and Wulf (1999) reported differences in a balance task between two groups of participants who saw the same visual feedback display but received instructions that directed attentional focus either internally or externally. However, Shea and Wulf also found that the inclusion of the visual feedback display improved performance relative to a no-feedback condition, even when the less effective IF instructions were provided. It was concluded that the visual display "might have served as a remote focal point that generally tended to induce an external focus, independent of the focus instructions" (Wulf, 2007: 5).

In summary, the nonspeech motor learning literature suggests that the nature of verbal cues provided has the potential to influence learning outcomes in biofeedback treatment. Specifically, previous results suggest that learning might be maximized if explicit articulator placement cues were replaced with cues encouraging an external focus of attention. This is a counterintuitive suggestion from a clinical standpoint, where there is a strong belief that placement cues are vital for learners to identify an appropriate articulatory configuration (e.g., Shuster et al., 1992). Because this could be a case where the optimal conditions for learning differ between speech and nonspeech tasks, it is essential to conduct direct investigation of the influence of attentional focus cues on outcomes in biofeedback intervention for speech disorders.

In the present study, children with residual /r/ misarticulation received eight weeks of visual-acoustic biofeedback intervention. All participants saw the same visual-acoustic biofeedback display during treatment. However, participants were assigned to different focus conditions defined by the nature of instructions and verbal cues provided. Participants in the internal focus (IF) condition received explicit articulator placement cues, while participants in the external focus (EF) condition received only cues pertaining to the feedback display. In light of previous research reporting the efficacy of visual-acoustic biofeedback intervention for residual speech errors (Shuster et al., 1992; Shuster et al., 1995, McAllister Byun and Hitchcock, 2012), it was hypothesized that both groups would exhibit improvement in the perceptually rated accuracy of /r/ production over the course of therapy. Based on the results of previous studies in the nonspeech motor domain (Freedman et al., 2007; Shea and Wulf, 1999), it was hypothesized that effect sizes would be significantly larger in the EF condition, when both the visual display and the clinician's verbal cueing served to direct the participant's attention externally.

Methods & Procedures

Participants

The study initially enrolled 10 monolingual speakers of American English ranging in age from 6;8 to 13;3 (mean age 10;0). Seven participants were male, and three were female, a ratio consistent with previous descriptions of residual speech errors (Shriberg, 2010). One male participant (IF condition) dropped out in the 4th week of the study due to scheduling conflicts. Participants were recruited primarily by referral from their school or private speech-language pathologists. For inclusion, participants were required to score within one standard deviation of the mean for their age on the "Auditory Comprehension" subtest of the *Test of Auditory Processing Skills-3rd Edition* (*TAPS-3,* Martin and Brownell, 2005)[1] and exhibit no more than two sounds other than /r/ in error on the *Goldman-Fristoe Test of Articulation-2nd Edition* (*GFTA-2,* Goldman and Fristoe, 2000). Participants were required to pass a pure-tone hearing screening at 20 dB HL, and to exhibit no gross structural or functional deficits in an informal administration of the Oral Mechanism Screening Examination, 3rd Edition (St. Louis and Ruscello, 2000) by a certified speech-language pathologist. Participants were required to have received intervention targeting /r/ for at least five months prior to enrollment, but they were asked to discontinue outside /r/ treatment while participating in the study. Per parent report, the duration of participants' previous intervention targeting /r/ ranged from five to 120 months (median duration 33 months). Parents were additionally asked to report what sounds other than /r/ were previously targeted in their child's therapy, but no other information about the nature of previous intervention (e.g., intensity, treatment methodology) is available. Participant details are summarized in table 1.

A second set of inclusionary criteria pertained to participants' pre-treatment accuracy in /r/ production. Stimulability was assessed with a standard protocol (Miccio, 2002), and children were excluded if their stimulability exceeded 30% for more than one variant of /r/. Participants were also required to score ≤30% correct on an 84-item probe eliciting /r/ at the word level in various phonetic contexts. The full probe measure, along with details of how the list was constructed, is available in On-line Supplement A.

Design

The study followed a single-subject experimental design with baseline, treatment, and maintenance phases. In the baseline phase, the 84-word /r/ probe was elicited in at least three sessions over a minimum of 1.5 weeks. Participants then received visual-acoustic biofeedback intervention in two 30-minute sessions per week for eight weeks. During the treatment phase, /r/ production accuracy was evaluated with a 36-item subset probe administered at the start of every third session. Subset probes included all words targeted in treatment, as well as untreated words and /r/ variants. At the conclusion of the study, the 84-

[1]One participant, "Johnny," received a standard score of 60 on the *TAPS-3* "Auditory Comprehension" subtest. However, in light of parent report that Johnny had no difficulty with listening tasks in home and classroom settings, he was re-tested using the "Elaborated Phrases and Sentences" subtest of the *Test for Auditory Comprehension of Language*, where he scored within the average range (standard score of 13). Johnny was thus included in the study on the grounds that his first score may not have been representative of his true auditory comprehension abilities.

word /r/ probe was again elicited in three maintenance sessions over approximately 1.5 weeks (mean = 11 days, SD = 5 days).

The study was designed to establish experimental control by collecting multiple baselines across participants and behaviors. The duration of the baseline period was increased by one session as successive children enrolled in the study, up to a cap of five sessions. The across-behaviors manipulation involved staggered introduction of distinct targets, i.e. different variants or allophones of /r/, across at least two phases of treatment. In Phase 1, one selected /r/ variant was treated while other variants were held at baseline; in Phase 2, a different variant was treated while the Phase 1 target was probed but not treated. The switch to Phase 2 was determined using a combined time and performance criterion. Whenever a participant met the performance criterion laid out below in "Treatment Protocol," a new target was introduced. If the performance criterion had not been satisfied after four weeks, Phase 2 was initiated by the time-based criterion. Targets were selected in an individualized fashion from among variants exhibiting a low, stable level of accuracy across previous probes. Whenever possible, targets alternated across treatment phases between vocalic variants of /r/ (i.e., syllabic /r/ and rhotic diphthongs) and consonantal variants (i.e., onset /r/). This was intended to minimize the potential for generalization between treatment targets, because treatment gains on vocalic variants of /r/ tend not to transfer spontaneously to consonantal /r/, or vice versa (Curtis and Hardy, 1959; McAllister Byun and Hitchcock, 2012; Preston et al., 2013). Within a treated category, three words were selected to be practiced in treatment, and three were left untreated to evaluate generalization. A complete list of words targeted for each participant is provided in table 2.

At the start of the study, participants were assigned to receive either internal focus (IF) or external focus (EF) cues during biofeedback treatment. Pseudo-random assignment was used so that each focus condition featured a balanced representation of children with respect to age, initial severity level, and duration of previous treatment. Participants were intended to remain in a single focus condition throughout the study. However, if a participant's within-session accuracy, as rated online by the treating clinician, did not exceed 20% for at least two sessions, the treatment condition was switched when the Phase 2 targets were introduced. This provision was included for ethical reasons, so that participants were not obligated to continue a treatment with a low likelihood of success. All methods described here were approved by the Institutional Review Board at Montclair State University. Parental consent and child assent were obtained for all participants.

The present study was structured to meet What Works Clearinghouse (WWC) standards for single-subject experimental design (Kratochwill et al., 2013). In particular, the use of nested across-subjects and across-behaviors comparisons meets the WWC requirement for a minimum of three opportunities to demonstrate an experimental effect at different points in time. In addition, the inclusion of a minimum of three points of data collection in each phase of the study meets WWC standards "with reservations"; a minimum of five observations per phase would be needed to satisfy the standards in full.

Treatment Protocol

All treatment in the present study was provided by a single trained graduate student in speech-language pathology, who was supervised by a single certified speech-language pathologist. During treatment, participants in both EF and IF conditions viewed a visual representation of the acoustic signal of their speech generated by the Real-Time LPC function of the Sona-Match Module of the Computerized Speech Lab (CSL; KayPentax). Figure 1 provides an example of this visual display. The solid shape labeled *LPC spectrum* represents the formants of the participant's speech, generated in real time. The line labeled *Template* is a trace taken from the LPC spectrum of a typical speaker's perceptually correct production of /r/. Because formant locations are influenced by the size and shape of the speaker's vocal tract, samples of perceptually correct /r/ were elicited from speakers representing a range of ages and body sizes, and participants were paired with the template judged to represent the best match for their estimated vocal tract size. In visual-acoustic biofeedback intervention for /r/, the treating clinician calls attention to a salient acoustic hallmark of /r/: the low height of the third formant, F3 (Delattre and Freeman, 1968; Hagiwara, 1995). F3 may lower to a point where it appears to merge with the relatively high second formant, F2 (Boyce and Espy-Wilson, 1997); this can be seen in figure 1, where the /r/ template features a single peak representing both F2 and F3.

In the first three treatment sessions, participants received extended instructions that lasted for 5–10 minutes prior to the initiation of treatment trials. Children in both focus conditions received a standard training protocol designed to familiarize them with the biofeedback technology (McAllister Byun and Hitchcock, 2012). Additional instructions introduced key concepts to be invoked in subsequent treatment sessions. Instructions differed across IF and EF conditions, but they were balanced for overall length (number of words) and complexity (Flesch-Kincaid grade level). To maintain a manageable cognitive load, only one major cue was introduced per session.

In the IF condition, extended instructions featured line drawings depicting tongue shapes associated with correct production of /r/. Each of the three instructional sessions cued one major component of tongue placement for /r/: tongue root retraction (Adler-Bock et al., 2007; Klein, McAllister Byun, Davidson, and Grigos, 2013), lateral tongue bracing (Bacsfalvi, 2010), and anterior tongue elevation. Explicit instructions to adopt a retroflex or bunched tongue shape were not provided (Klein et al., 2013). In the EF condition, extended instructions and additional images of biofeedback displays were used to help participants understand how to use the visual-acoustic display to achieve a better /r/ sound. The first session emphasized the idea that learners could move the LPC spectrum around by changing the sound of their speech; the second session provided additional practice in discriminating LPC spectra associated with correct versus incorrect /r/ sounds; and the third session encouraged participants to identify incremental changes in the location of the formants and shape those changes into a more accurate /r/ sound. No instructions about the articulatory properties of /r/ were provided in the EF condition.

After the instructions in sessions one through three, and at the start of all subsequent sessions, participants completed a period of roughly five minutes devoted to biofeedback "free play." They were encouraged to try out any manipulations they liked while observing the LPC display. Participants subsequently practiced /r/ at the word level in 60 trials, where each of three individually selected target words was elicited in four blocks of five trials. The order of blocks was randomized. Blocks were preceded by one focusing cue and followed by one piece of qualitative feedback, which differed across the EF and IF conditions. In the EF condition, participants received only cues that referred to the LPC spectrum (e.g., "Focus on making the wave match the red line"). In the IF condition, cues evoked the tongue placement instructions provided in the first three treatment sessions. Participants in the IF condition could also receive cues pertaining to the biofeedback display, but these were always paired with an articulatory cue. Feedback was of the qualitative or *knowledge of performance* (KP) type, and it was consistent with the participant's focus condition. Examples included "Try to open your jaw a bit wider" for IF and "You got the third bump closer to the second bump that time" for EF. Audio playback was provided after 50% of blocks, randomly scheduled. After audio playback, the participant was asked to report which trial he/she thought was most accurate, and the clinician offered feedback on the participant's self-rating. Audio playback was included to facilitate the skill of perceptually judging the accuracy of /r/ sounds, so participants would not be wholly dependent on visual feedback.

To encourage generalization of new speech-motor skills to a more naturalistic context, a biofeedback fading schedule was enacted if participants achieved a high level of accuracy within practice. Biofeedback was initially provided in 100% of trials. If a participant achieved 70% accuracy in a session, biofeedback frequency was reduced to 80% of trials (four out of five trials per block) in the following session. If 70% accuracy was again attained, biofeedback was reduced to 60% and then 40% of trials. If a participant demonstrated at least 80% accuracy with 40% biofeedback frequency, the performance criterion was met and a new /r/ variant was introduced. Biofeedback withdrawal, which was achieved by holding a barrier in front of the computer screen, was randomly scheduled within each block.

Treatment Fidelity

Fifteen percent of all sessions were reviewed to evaluate fidelity to the stated treatment protocol (Kaderavek and Justice, 2010). To measure fidelity, the audio record of a treatment session was reviewed by research assistants not involved in the treatment portion of the study. These raters completed a checklist to verify the following aspects of the study design: (1) each block of five trials was preceded by a reminder cue, (2) this cue was consistent with the child's treatment condition (IF/EF); (3) each block consisted of precisely five trials; (4) feedback or other interruptions did not occur within a block; (5) audio playback was presented in 50% of blocks; (6) qualitative (KP) feedback was provided after each block, and (7) the feedback provided was consistent with the child's focus condition.

Measurement

Binary perceptual ratings of /r/ sounds elicited in probe measures were collected from non-specialist listeners who were recruited through the online crowdsourcing platform Amazon Mechanical Turk (AMT; e.g., Mason and Suri, 2012). Sound files of whole words were presented and responses collected using the online experiment presentation platform Experigen (Becker and Levine, 2010). This use of AMT was approved by the Institutional

Review Board at New York University, and participants and their parents gave consent for sound files to be shared with external listeners in an anonymized fashion for rating purposes. Although recent years have seen extensive use of AMT by researchers in psychology (e.g., Paolacci, Chandler, and Ipeirotis, 2010) and linguistics (e.g., Gibson, Piantadosi, and Fedorenko, 2011), this study represents a novel application of AMT to evaluate the accuracy of recorded speech. The validity of AMT listeners' ratings relative to experienced listeners' ratings was rigorously vetted in a companion study (McAllister Byun, Halpin, & Szeredi, 2015). Since aggregating across multiple individuals is a way to reduce noise in rating tasks (Ipeirotis et al., 2014), that paper asked how many nonexpert AMT listeners would be needed to equal the performance of the "industry standard" in which responses are aggregated across three experienced listeners. A bootstrap analysis found that the "industry standard" level of agreement with an expert listener gold standard was achieved when responses were aggregated across samples of at least nine AMT listeners. Therefore, the task on AMT was structured to collect nine unique listeners' ratings of each item. Sometimes fewer responses were collected for a given item due to data loss (e.g., sound file failed to play). Items with eight unique listener ratings were considered adequate for inclusion in the analysis; items with seven or fewer ratings were added to later blocks to collect additional ratings.

Several steps were implemented to train listeners and verify that they maintained attention to the task. These methods are described in detail in On-line Supplement C, as well as in McAllister Byun, Halpin, & Szeredi, 2015. After training and passing an eligibility test, listeners rated the accuracy of the /r/ sound in each word by clicking "correct" or "incorrect." Raters saw the target word in standard English orthography and could listen to each word up to three times. Files were presented in blocks of 200. Raters could complete multiple blocks, but after five blocks, they were required to pass the eligibility test again. A total of 249 unique raters completed at least one block. The average number of blocks completed per rater was 2.4 (range: 1–46). Each block contained 20 attentional catch trials, which were judged by the experimenters to be unambiguously correct or incorrect. If a participant did not score above chance on the catch trials in a block, the block was discarded.

Each item was rated by at least eight unique listeners. Interrater agreement was calculated across the set of listeners scoring a given token. For the full set of data, agreement was an acceptable 80.7%. Agreement was poorer for consonantal variants of /r/ (76.1%) than for vocalic variants (85.7%). To correct for this slightly elevated level of interrater variability, each item was assigned a single accuracy rating reflecting the mode across eight or more listeners. In the event of a tie in an even-numbered sample of raters, the lower modal value (i.e., 0) was reported.

Analyses

Following the traditional method of analysis for single-subject experimental studies, results for each participant were analyzed for evidence of a treatment effect via visual inspection (Kratochwill, Levin, Horner, and Swoboda, 2014). However, visual analysis has been criticized for low interrater reliability (e.g. Brossart, Parker, Olston, and Mahadevan, 2006). It has been suggested that the scientific credibility of single-subject experimental research could be enhanced by incorporating converging evidence from quantitative outcome measures, including effect sizes and hypothesis tests or calculation of confidence intervals (Kratochwill and Levin, 2014). The present study adopted a multi-pronged analytical approach in which visual inspection was supplemented with individual effect sizes, as well as a logistic mixed model examining across-subjects effects. By triangulating across these different outcome measures, we can arrive at an overall impression of response to treatment that is more robust than any single measure.

Effect sizes were calculated for each individual by comparing /r/ productions elicited in single-word probes administered during the true baseline phase (prior to the initiation of any treatment) and the post-treatment maintenance phase. Effect sizes were standardized using Busk and Serlin's d_2 statistic (Beeson and Robey, 2006), which pools standard deviations across baseline and maintenance periods in order to reduce the number of cases where effect size cannot be calculated due to zero variance at baseline. The present study follows Maas and Farinella (2012) in setting 1.0 (i.e., the difference between pre- and post-treatment means exceeds the pooled standard deviation) as the minimum standardized effect size that will be considered clinically relevant.

Across-subject comparisons were computed in a logistic mixed model, which can be used when the dependent variable is binary and the independent variables include both fixed/repeatable and random/non-repeatable factors (e.g., McAllister Byun and Hitchcock, 2012). The model was structured with reference to Rindskopf and Ferron's (2014) guidelines for multilevel modeling of single-subject data. As in the calculation of effect sizes, the mixed model compared the perceptually rated accuracy of productions during the true pre-treatment baseline phase against the true post-treatment maintenance phase. Both treated and untreated words and /r/ variants were represented in all probe measures. The dependent variable was the perceptual accuracy rating assigned to each word-level /r/ production, determined based on the mode across at least eight nonexpert listeners recruited through AMT. A random effect of participant was included to adjust for the fact that individual children could have different initial levels of accuracy or differing rates of response to therapy.

A fixed effect of treatment phase evaluated whether /r/ sounds elicited during baseline versus maintenance periods, aggregated across participants, differed in the likelihood of being rated 'correct.' Another predictor examined whether the likelihood of a 'correct' rating differed across treated and untreated variants of /r/. This predictor is of primary interest in its interaction with the fixed effect of treatment phase, indicating whether treated and untreated variants differed in the likelihood of change over the course of treatment. The model also included a fixed effect of direction of attentional focus cueing, evaluating whether participants' progress in biofeedback treatment was modulated by the internally- versus externally-orienting nature of verbal cues provided. To address the complication whereby children who demonstrated minimal response during the first four weeks of treatment could undergo a change in attentional focus cue condition, participants were coded into three attentional focus conditions: IF, EF, and mixed. First-order interactions of direction of focus cues with treatment phase and with the treatment status of a particular /r/ variant were included in the model, as well as a three-way interaction among these three factors.

Results

Treatment Fidelity

As noted above, recordings of 15% of all treatment sessions were reviewed by research assistants to check fidelity to the stated treatment protocol. The primary deviations from protocol involved the absence of a reminder cue or KP feedback; these inputs were provided preceding 91% and following 85% of the blocks reviewed. When not omitted, however, cues and feedback were consistent with the child's assigned focus condition in 99% of cases. An additional source of deviation occurred in the form of within-block interruptions or feedback, which were observed in approximately 3% of blocks. Full details of the fidelity check can be found in On-line Supplement B.

Individual results: Visual inspection

In the multiple-baseline graphs that follow, each child is represented by two to three boxes corresponding with the /r/ variants targeted over successive phases of treatment. In each box, the treated interval is shaded gray. The values plotted represent the percentage of words in the treated category that were rated correct based on the mode across at least eight blinded listeners. Here and elsewhere, "words in the treated category" includes both treated words and words containing the same /r/ variant that were not specifically practiced in treatment.

For convenience, the single-subject graphs have been grouped into three sets of three participants. The first group includes three participants who underwent a change in attentional focus over the course of the study as a consequence of minimal response to treatment. The second and third groups each include three children who received treatment exclusively in the IF or EF condition, respectively.

Figure 2 depicts the response to treatment of the three participants who underwent a change in attentional focus over the course of the study. The two youngest participants, Johnny and Maya, did not show an increase in accuracy on treated targets in the first four weeks of the study, even within the treatment setting while using the biofeedback device. Johnny's treatment started in the EF condition, while Maya's began in the IF condition. In accordance with the protocol laid out above, both underwent a switch in direction of attentional focus at midpoint, to evaluate the possibility that different cues might facilitate a greater degree of progress. Neither showed a sustained pattern of improvement following this switch. A third participant, Benny, underwent a change in attentional focus condition due to an error in the implementation of the experimental protocol. While it was originally believed that Benny met the criteria for a switch from EF to IF due to lack of progress, subsequent review revealed that the treating clinician had assigned accuracy scores of 40–50% in the last two treatment sessions in Phase 1, a notable increase. However, neither treatment condition yielded gains that generalized to treated targets elicited without biofeedback.

Figure 3 depicts the treatment response of the three participants who received intervention in the IF condition, in which biofeedback was accompanied by articulator placement cues. The first participant, Isabel, made visually salient gains for the Phase 1 target (rhotic diphthong /ɔr/), but she did not make progress that generalized to a context without biofeedback on the Phase 2 target (unstressed /ɚ/). By contrast, Steven did not show improvement on word probes for the Phase 1 target (singleton /r/ with back vowels), but he clearly responded to treatment of the Phase 2 target (stressed /ɝ/) and progressed to a third phase targeting labial clusters. In Phase 3, he again showed a marked increase in the accuracy of the treated category, although these gains were not maintained as strongly as in the case of the Phase 2 target. The third participant, Brendan, was judged to produce highly accurate /r/ sounds during biofeedback treatment, but generalization to words produced without biofeedback was minimal.[2]

Figure 4 depicts the response to treatment of the three participants who received treatment in the EF condition, i.e. with no articulator placement cues. Robert showed limited progress during Phase 1, but both the Phase 1 and Phase 2 targets (rhotic diphthongs /ɛr/ and /ɔr/) showed improvement during the second phase of treatment. However, these gains were small in magnitude and intermittently sustained. The second participant, Erin, was judged to produce consonantal /r/ with relatively high accuracy in the baseline phase. Therefore, two vocalic variants (rhotic diphthong /ɪr/ and stressed syllabic /ɝ/) were targeted in Phases 1 and 2. Erin reached and sustained near-ceiling-level accuracy for both of these targets. Unfortunately, within-subject control was compromised by carryover between the two targets, which both showed increasing accuracy during both phases of treatment. After the vocalic variants reached ceiling-level accuracy, Erin received a third phase of treatment targeting a consonantal variant (singleton /r/ before back vowels), but no generalization gains were observed for this target. Phase 1 and 2 probe data are not available for this target because consonantal variants were not initially considered as treatment targets for Erin and therefore were excluded from her individualized within-treatment probe list. The final participant, Joseph, showed rapid progress within the treatment setting for his Phase 1 target, labial clusters. The duration of Phase 1 was shortened because he met the performance criterion (80% accuracy with 40% biofeedback frequency) for advancement to a new target. However, these gains did not generalize to word probes elicited without biofeedback until Phase 2, suggesting that a more stringent criterion for advancement might have been more appropriate. Joseph showed strong gains following the onset of treatment for the Phase 2 target (rhotic diphthong /ɪr/), but these gains were not robustly maintained through all post-treatment maintenance sessions.

Individual Results: Effect Sizes

Table 3 provides a summary of standardized effect sizes across all participants in treatment phases 1, 2, and 3 (where applicable). Effect sizes were calculated for both the treated variant in each phase (including both treated and untreated words) and untreated variants. Standardized effect sizes that exceed the minimum clinically significant value ($d_2 > 1.0$) are in bold. Table 3 shows that six out of nine participants achieved a clinically significant effect size for at least one treated target. Of these six, three received treatment in the IF condition and three in the EF condition. Independent of attentional focus condition, there was a

[2]Based on the standardized effect sizes reported in Table 3, Brendan is classified as a child who exhibited a clinically significant response to treatment. In this case, the standardized effect size is somewhat inflated by low variance, as indicated by a small raw difference between the mean percentage of items rated correct in maintenance versus baseline intervals. Taken in combination with inspection of visible trends, the calculated effect sizes indicate that Brendan did respond to treatment, but the magnitude of change over the course of the study was limited.

significant correlation between age in months and the mean standardized ES across all treated targets (Spearman's ρ = .79, $p = .01$).

Mixed model results

In the logistic mixed model described above, treatment phase was a significant predictor of /r/ production accuracy ($\beta = 0.39$, $z = 3.4$, $p < 0.001$). In this case, the positive coefficient indicates that /r/ sounds produced during the maintenance phase were more likely to be rated perceptually correct than /r/ sounds produced during the baseline phase. The treatment status of a given /r/ variant (treated versus untreated in a given individual) was also a significant predictor of perceptually rated accuracy of /r/ in words ($\beta = -1.2$; $z = -5.3$; $p < 0.001$). It initially seems surprising that this factor has a negative coefficient, indicating that /r/ variants never targeted in treatment were more likely overall to be rated perceptually accurate than treated variants. Recall, however, that /r/ variants that received high perceptual accuracy ratings during the initial evaluation were not eligible for selection as treatment targets. Furthermore, there was a significant interaction of treatment phase and treatment status ($\beta = 1.0$; $z = 3.2$; $p < 0.01$), reflecting the fact that treated variants were more likely to change in perceptually rated accuracy over the course of treatment than untreated variants. There was also a main effect of direction of attentional focus condition (EF versus mixed: $\beta = -2.6$; $z = -3.8$; $p < 0.001$), indicating that /r/ sounds produced by the participants who underwent a change in direction of attentional focus were overall less likely to be rated perceptually correct than /r/ sounds produced by participants who received only EF- or IF-orienting cues. This is unsurprising, since a change in attentional focus condition was introduced only when a participant failed to respond to treatment. The comparison between EF and IF attentional focus conditions was not significant (β =0.43; $z = .66$; $p = 0.51$). There were no significant interactions between attentional focus condition and treatment phase (EF versus IF: $\beta = .22$; $z = 1.4$; $p = 0.16$; EF versus mixed: $\beta = -0.15$; $z = -0.62$; $p = 0.53$), meaning that the likelihood of progress over the course of treatment did not differ as a function of the nature of attentional cues provided. There were no other significant effects or interactions in the model. Complete regression results are reported in On-line Supplement D.

Discussion

This study was undertaken with two aims: to collect systematic evidence on the efficacy of visual-acoustic biofeedback intervention for residual /r/ errors, and to evaluate the influence of internally versus externally orienting attentional cues during biofeedback treatment. We begin the discussion by addressing our most basic experimental question: Do the results of this single-subject study support visual biofeedback as an effective form of intervention for children whose errors have not responded to traditional forms of intervention? We then go on to examine the effect of EF versus IF cue conditions on the magnitude of response to biofeedback treatment.

Effects of biofeedback treatment

The logistic mixed model, in which outcomes were pooled across participants for analysis, suggests a positive impact of biofeedback intervention. The significant effect of treatment phase in the logistic mixed model indicates that /r/ words produced after eight weeks of biofeedback intervention were more likely to be rated perceptually correct than the same words produced at baseline. Evidence that these gains can be attributed to the treatment rather than maturation or external influences is contributed by the significant interaction between treatment phase and treatment status of /r/ variants, which indicates that /r/ variants targeted in treatment were more likely to undergo a change in perceptually rated accuracy than untreated variants. When we examine individual effect sizes, we obtain corroborating evidence in support of the efficacy of biofeedback intervention: six out of nine participants showed clinically meaningful progress (ES > 1.0) on at least one treated /r/ variant. Visual inspection indicated that gains generally occurred following the onset of biofeedback treatment, supporting the hypothesis that the observed changes in accuracy were causally linked to the application of treatment.

The present findings provide a preliminary suggestion that the effectiveness of visual-acoustic biofeedback intervention may differ as a function of age. All but one of the participants aged eight and above showed significant gains in accuracy on at least one /r/ variant. By contrast, the two children aged seven and below showed no sustained gains either within treatment or on word probe measures. As a post-hoc measure, we examined the correlation between age in months and mean standardized ES across treated targets and found it to be significant (Spearman's $\rho = .79$, $p = .01$). Previous research has shown that it is possible for children younger than seven to make significant progress in biofeedback treatment (e.g., McAllister Byun and Hitchcock, 2012). However, the present results suggest that a successful response to biofeedback intervention may be more likely when participants are slightly older. On the other hand, one of the participants who showed no response to treatment, Johnny, also demonstrated questionable performance on the initial evaluation of auditory comprehension (see footnote 1). We cannot rule out the possibility that subclinical difficulties with auditory perception might have played a role in his lack of response to treatment. Follow-up research should examine the interaction between age, auditory comprehension, and response to biofeedback intervention in a more systematic fashion.

Previous research has reported that gains made within the context of biofeedback treatment do not automatically generalize to a context in which biofeedback is not available (e.g., Fletcher, Dagenais, and Critz-Crosby, 1991; Gibbon and Paterson, 2006; McAllister Byun and Hitchcock, 2012). The present study provides corroborating results: participants such as Brendan (IF condition) and Joseph (EF condition) showed considerable progress within the treatment setting but were less successful in transferring these gains to other contexts. Promptly reducing the frequency of biofeedback may enhance the likelihood of successful generalization; see discussion in Hitchcock and McAllister Byun (2014).

Effects of internal versus external focus cues

The mixed logistic model revealed no significant differences between participants who received internally-focusing versus externally-focusing cues, either with respect to overall accuracy or in the magnitude of improvement over the course of treatment. It is possible that this null result can be attributed to the low experimental power of the analysis, although the data do not show a meaningful trend in either direction. Inspection of individual results also fails to reveal a clear advantage for one attentional focus condition over the other: out of the six participants who attained a standardized ES greater than 1.0 on at least one treated target, three had been randomly assigned to the EF condition and three to the IF condition.

Since the present results do not show an advantage for participants receiving EF cues, it might be supposed that an external direction of attentional focus is not a key component in the efficacy of biofeedback treatment. Taking this result in the context of previous research, though, we favor a different interpretation. Chiviacowsky, Wulf, and Ávila (2013) have argued that when rich visual feedback is provided during a motor learning task, attention may be drawn externally even if IF instructions are provided (see also Wulf, 2007; Wulf and Shea, 1999). Thus, one possible interpretation is that the presence of biofeedback in itself induced an external focus of attention, beyond which verbal instructions did not provide additional benefit. While the present results do not provide direct evidence that external focus of attention makes an active contribution to the effectiveness of biofeedback, they also are not incompatible with this hypothesis.

Furthermore, the present finding of comparable gains on treated targets across IF and EF conditions is in itself a result of some clinical and theoretical significance. In the EF condition, participants like Erin and Joseph acquired correct /r/ through biofeedback treatment in the absence of any explicit articulatory cues. Most practitioners report adopting an articulation-oriented approach to intervention for speech sound errors (Brumbaugh and Smit, 2013), suggesting that they regard explicit placement cues as an important ingredient for progress in treatment. The use of placement cues is especially common when errors are judged to have an articulatory or motoric origin; /r/ misarticulation is perhaps the foremost example of this category. The present study supports other recent research (e.g., Rvachew and Brosseau-Lapré, 2012) in finding that correct production can be achieved through intervention emphasizing the acoustic or auditory properties of a target sound, even in the absence of explicit articulator placement cues.[3] Our results are also compatible with theoretical models arguing that speech targets are primarily acoustic rather than articulatory in nature (e.g. Guenther, Hampson, and Johnson, 1998; Perkell, 2012).

Lastly, from a clinical perspective, the present findings can be viewed as a ‘best of both worlds’ scenario. Experienced clinicians generally have a set of cueing strategies that they have found to be facilitative in past practice, and it is unlikely that clinicians who choose to incorporate biofeedback into their practice would want to do away with these cues altogether. Our results provide no indication that explicit articulator placement cues impede progress in visual-acoustic biofeedback treatment. On the other hand, these results do suggest that a clinician using biofeedback methods is also free to refrain from providing explicit placement cues in contexts where they are judged unlikely to be facilitative. This has particular relevance in the context of treatment for /r/ misarticulation. It is known that typical speakers differ in the tongue postures they adopt to produce /r/ (e.g., Delattre and Freeman, 1968; Zhou et al., 2008), and there is a lack of evidence to indicate which of these postures

[3]Given that participants in the present study had all received intervention in other settings in the past, we cannot rule out the possibility that they were using their memory of articulator placement cues provided previously to facilitate their learning during biofeedback. If this were the case, it would tend to support our hypothesis that external direction of attentional focus is facilitative in speech-motor learning, given that the participants were apparently unable to benefit from these placement cues until they no longer constituted the primary emphasis of treatment.

should ideally be targeted during /r/ intervention (see discussion in McAllister Byun, Hitchcock, and Swartz, 2014). It is thus useful to have the option to provide cues and feedback based on the acoustic characteristics of /r/, either via biofeedback or auditory models, and make no commitment to one articulatory variant or the other.

Conclusions

This study addressed two experimental objectives: to collect systematic evidence on the efficacy of visual-acoustic biofeedback intervention for residual /r/ errors, and to evaluate the influence of internally versus externally orienting attentional cues during biofeedback treatment. A single-subject experimental design with multiple baselines across participants and behaviors was used. To draw more robust conclusions, outcomes were evaluated using several distinct analytical methods (visual inspection, individual effect sizes, logistic mixed modelling). These analyses offered evidence supporting the efficacy of biofeedback intervention, albeit with variability across individual participants. Six out of nine participants showed sustained improvement on at least one treated target after eight weeks of treatment, despite a previous history of no successful response over months or years of traditional treatment. Of these responders, three received treatment in the internal focus condition and three in the external focus condition. The mixed model also revealed no significant difference between the two direction of focus cue conditions. These results are consistent with previous findings that visual biofeedback may be sufficient to establish an external direction of attentional focus, independent of the nature of any verbal cues provided. The finding that explicit articulator placement cues were not necessary for progress in treatment has implications for intervention practices for speech sound disorders in children.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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What this paper adds

Nonspeech motor research suggests that motor skill acquisition is facilitated when the learner maintains an external direction of attentional focus during practice. However, there is little research to indicate whether these findings also apply to speech-motor learning in individuals with speech sound delay or disorder. An external direction of attention can be adopted in visual biofeedback intervention, but in practice, biofeedback treatment is often accompanied by articulatory cues that encourage an internal direction of attentional focus.

In the present study, outcomes did not differ significantly across participants who received biofeedback with externally orienting cues and participants who received biofeedback with internally orienting articulator placement cues. These results are consistent with previous findings that biofeedback may be sufficient to establish an external attentional focus, independent of the nature of verbal cues provided. The finding that explicit articulator placement cues were not necessary for progress in treatment has implications for intervention practices for speech sound disorders in children.

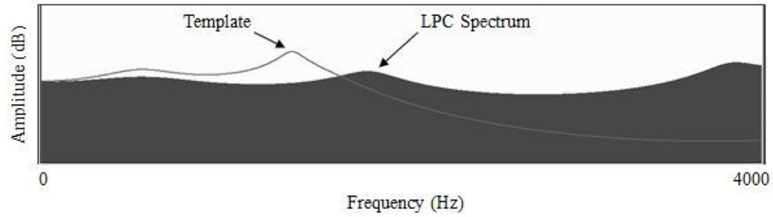

FIGURE 1.
Sona-Match display with LPC formant tracking and correct /r/ template. From McAllister Byun & Hitchcock (2012). Used with permission.

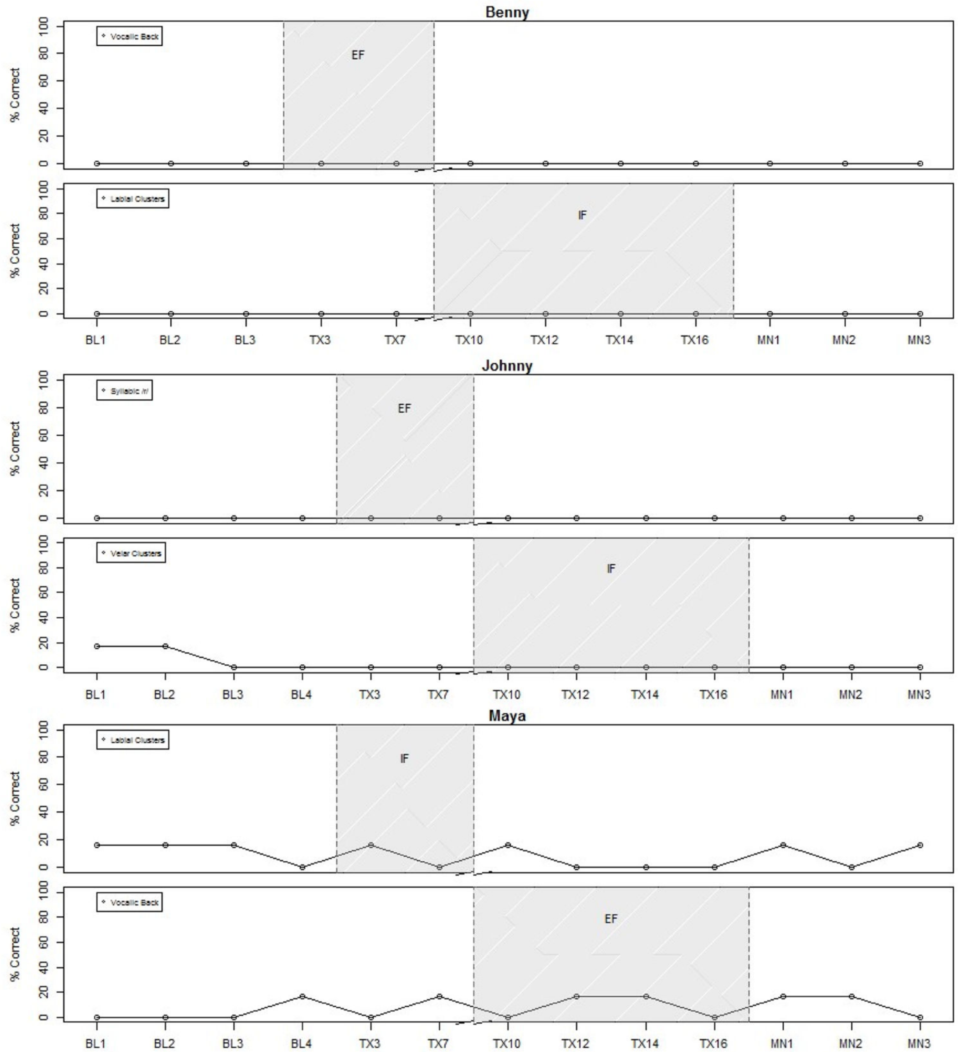

FIGURE 2.

Percent correct production of words in a treated category elicited in no-feedback probe measures. Participants in this figure underwent a switch in treatment condition due to a lack of progress in the treatment setting in Phase 1.

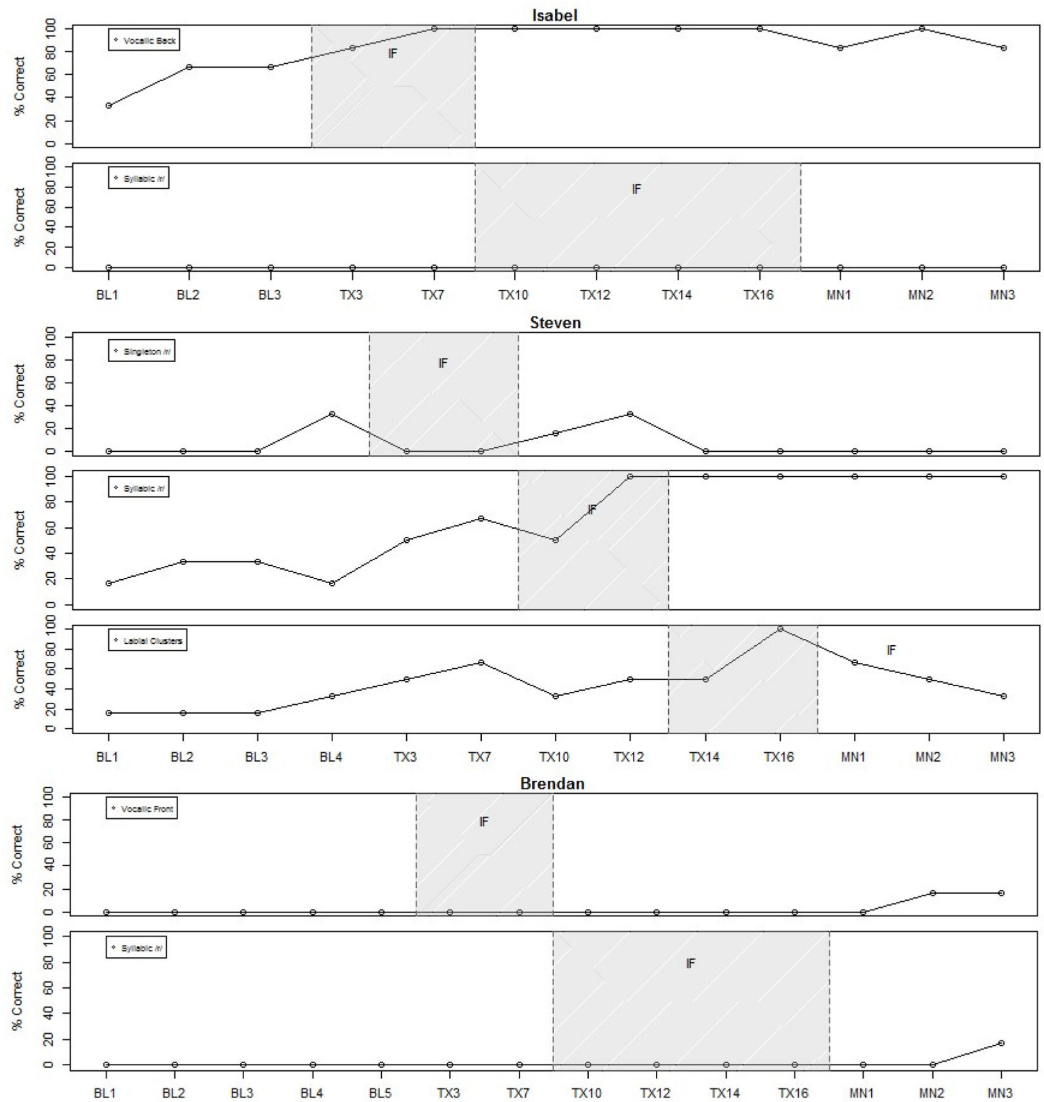

FIGURE 3.

Percent correct production of words in a treated category elicited in no-feedback probe measures: Internal focus condition.

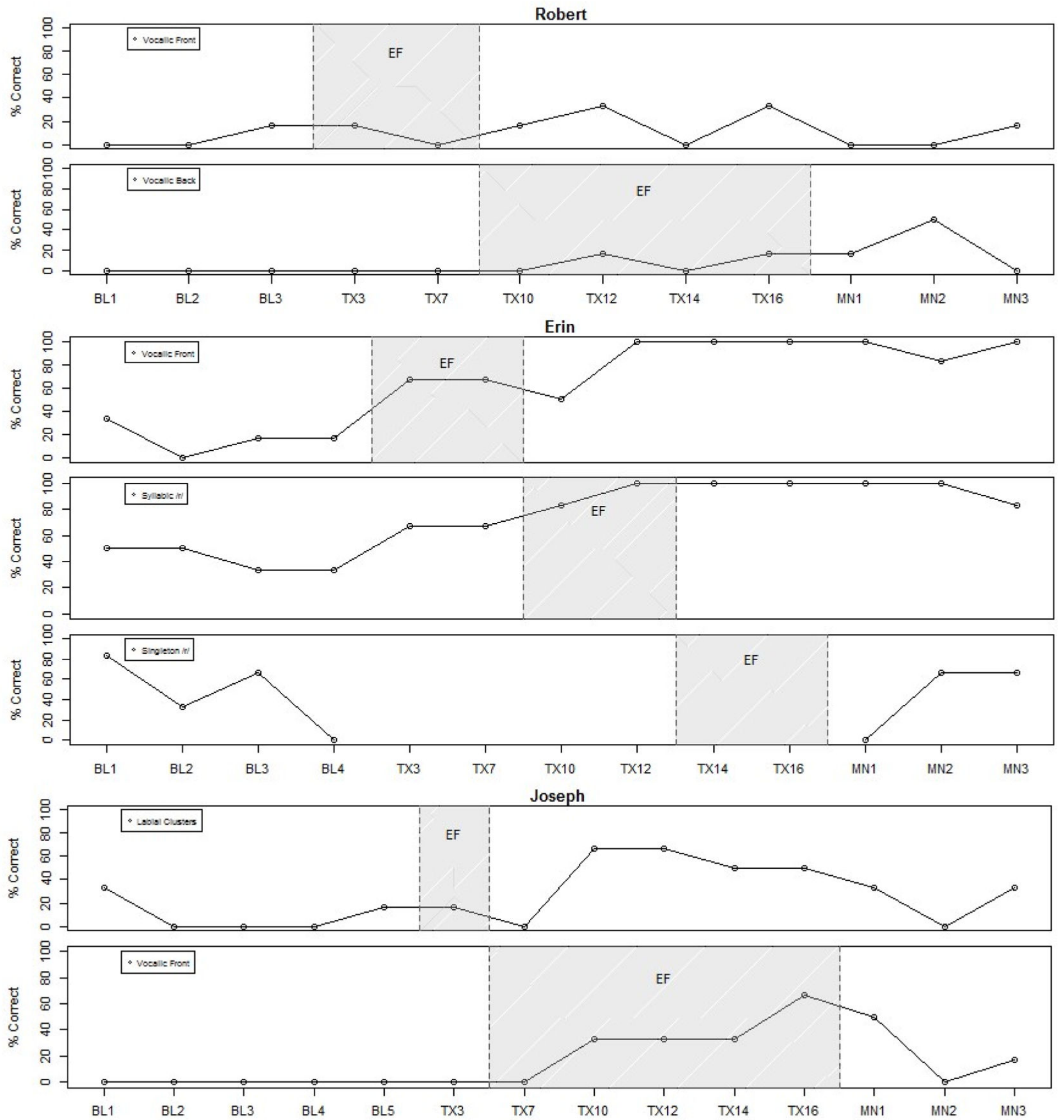

FIGURE 4.

Percent correct production of words in a treated category elicited in no-feedback probe measures: External focus condition.

TABLE 1

Characteristics of participants enrolled in the study.

Pseudonym	Sex	Age	Duration of previous treatment targeting /r/	Other targets previously treated
Benny	M	8;11	2 years	/s, ʃ, l/
Brendan	M	9;1	3 years	NONE
Chris *	M	13;1	2 years	/s, ʃ /
Erin	F	9;11	2.5 years	NONE
Isabelle	F	13;3	5 years	NONE
Johnny	M	6;8	1 year	/l/
Joseph	M	11;0	5 years	NONE
Maya	F	7;3	5 months	/θ/
Robert	M	9;0	5 years	/θ/
Steven	M	12;1	10 years	NONE

*Dropped out in the fourth week of the study.

TABLE 2

All target words treated for each participant.

Pseudonym	Initial Condition Assignment	Phase 1 Treated Words	Phase 2 Treated Words	Phase 3 Treated Words
Benny	EF	*scar, star, barn*	*froze, broom, brown*	
Brendan	IF	*tear, share, scare*	*turn, purr, sir*	
Erin	EF	*spear, clear, weird*	*turn, stir, worm*	*rug, rob, rock*
Isabel	IF	*fork, board, floor*	*sour, butter, ladder*	
Johnny	EF	*stir, purr, worm*	*cry, grape, crab*	
Joseph	EF	*break, bridge, fry*	*spear, year, weird*	
Maya	IF	*frame, fry, brick*	*board, store, fork*	
Robert	EF	*stare, chair, share*	*board, store, door*	
Steven	IF	*rug, rude, rob*	*nurse, sir, worm*	*broom, proud, froze*

TABLE 3

Standardized effect sizes (d_2) for all participants, grouped by focus condition. For treated variants, results are pooled across treated and untreated words. Effect sizes exceeding the minimum value considered clinically significant ($d_2 > 1.0$) are in bold.

Child	Focus	Phase 1 Treated	Phase 2 Treated	Phase 3 Treated	Untreated Vocalic	Untreated Consonantal
Maya	IF/EF	−0.2	0.8		N/A*	0.3
Johnny	EF/IF	N/A*	−1.1		−0.6	−0.1
Benny	EF/IF	N/A*	N/A*		N/A*	**1.6**
Isabel	IF	**2.2**	N/A*		0.9	**1.7**
Steven	IF	−0.6	**10.1**	**2.4**	**5.5**	0.7
Brendan	IF	**2.0**	**1.0**		**4.9**	**3.9**
Robert	EF	0.0	**1.2**		0.0	**1.8**
Erin	EF	**6.3**	**5.5**	−0.1	**2.3**	−1.2
Joseph	EF	0.7	**1.5**		**1.0**	0.9

* N/A: Effect size could not be computed due to zero variance. In the present study, all such cases were associated with unchanging 0% accuracy.