Active treatments for aprosodia secondary to right hemisphere stroke

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Abstract—This study investigates the effects of two mechanism-based treatments for expressive aprosodia. Three participants, two women and one man, had a right hemisphere cerebral infarction resulting in affective aprosodia with greater expressive than receptive deficits. Trained raters determined presence of aprosodia by judging participants’ performance on two emotional communication batteries. A single-subject design with replication across three participants was employed. Sentence production with the use of treated and nontreated emotions was measured during baseline and treatment phases. Sentences were scored for accuracy by a trained rater blind to time of testing and analyzed visually and statistically. Effect sizes calculated on the resulting data for each participant and treatment confirmed modest to substantial treatment effects for both treatments in all three participants. Because of a relative paucity of treatment studies investigating expressive aprosodia, these data are among the first to suggest that aprosodia may be amenable to behavioral treatments.

Key words: affect, aprosodia, behavioral treatment, emotional communication, prosody, rehabilitation, right hemisphere damage.

INTRODUCTION

Aprosodia has been defined as a disruption in the expression or comprehension of the changes in pitch, loudness, rate, or rhythm that convey a speaker’s emotional intent [1–2]. These expression and comprehension deficits can occur alone or in combination and both can have devastating effects on human interaction. Expressive aprosodia can leave the speaker unable to vary his or her voice to express joy, sadness, anger, and other common emotions. Even strongly emotional utterances may be spoken in a flat, unemotional tone [3]. In the receptive form of aprosodia, a person can have difficulty interpreting a speaker’s emotional intent [4], particularly when the semantic message conflicts with the speaker’s tone of voice [5].

Aprosodia and other behavioral signs and symptoms of right hemisphere damage following a stroke are notoriously difficult to reduce with behavioral treatment, in part because of the frequent presence of anosognosia, a pathological denial of impairment. A variety of treatments for

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aprosodia have been proposed [6]; however, few have been adequately tested. Two single-case studies of individuals with aprosodia [7–8] provide some limited behavioral data suggesting that aprosodia is amenable to treatment. Our previous study also showed a positive response to treatment for aprosodia [9]. Further data are needed, however, before clinicians can confidently apply behavioral treatments for this deficit.

This study determines the effects of two mechanism-based treatments for expressive aprosodia. The first treatment was an imitative paradigm motivated by the hypothesis that expressive aprosodia may result from impaired motor programming/planning of the vocal elements that constitute emotional prosody [10]. The other treatment was a cognitive-linguistic approach motivated by the hypothesis that aprosodia results from a deficit in a modality-specific nonverbal affect lexicon [11]. We used a single-subject ABAC design (described later in the “Experimental Design” section) replicated across three participants who had primarily expressive aprosodia. We hypothesized that both the imitative treatment and the cognitive-linguistic treatment would be active but that the imitative treatment would show greater relative activity. That prediction was based on the assumption that a primarily expressive aprosodia is induced by a programming/planning deficit.

METHODS

Participants

The three participants, two women and one man, were right-handed native English speakers who had had a right hemisphere stroke with resulting expressive aprosodia. The three participants were also assessed for the presence of receptive aprosodic deficits. All three had only mild receptive deficits and were therefore judged to exhibit a primarily expressive aprosodia. Table 1 lists relevant participant demographic information.

The presence and severity of receptive and expressive aprosodia were independently determined by four trained raters who judged each participant’s performance on two emotional communication batteries, the Florida Affect Battery [12] and an unpublished emotional expressive battery being developed by the Cognitive Neuroscience Laboratory at the University of Florida. The Florida Affect Battery, a battery of tests of receptive emotional communication, assesses the ability to identify spoken prosody as well as facial expression of emotional affects [12]. The expressive emotional communication battery has participants perform a series of subtests. The first three test the ability to imitate syntactic and emotional prosody, and the other three test production of syntactic and emotional prosody to command. These two batteries of emotional communication ability were administered before treatment began and again at the end of treatment. Table 2 lists scores for both batteries for all participants. Prior to beginning the study, all participants gave informed consent in accordance with a protocol approved by the Health Science Center Institutional Review Board at the University of Florida.

Table 1.
Subject demographic information.

<table>
<thead>
<tr>
<th>Subject</th>
<th>Age</th>
<th>Gender</th>
<th>Education Level</th>
<th>Occupation</th>
<th>Medications for Depression</th>
<th>Duration Postonset (mo)</th>
<th>Lesion Localization</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>57</td>
<td>F</td>
<td>RNA</td>
<td>Retired nurse</td>
<td>Zoloft 50 mg/day</td>
<td>9</td>
<td>Right hemisphere, parietal infarct with posterior temporal and posterior frontal involvement</td>
</tr>
<tr>
<td>2</td>
<td>52</td>
<td>F</td>
<td>High school</td>
<td>Housewife</td>
<td>Paxil 20 mg/day</td>
<td>6</td>
<td>Right hemisphere, involves centrum semiovale, extends to internal capsule, includes portions of striatum</td>
</tr>
<tr>
<td>3</td>
<td>49</td>
<td>M</td>
<td>High school</td>
<td>Truck driver</td>
<td>Prozac 10 mg/day</td>
<td>4</td>
<td>No imaging available, presence of dense left hemiplegia</td>
</tr>
</tbody>
</table>

RNA = registered nurse anesthetist.
tones and affect were less vibrant than before the stroke. She was residing in an assisted living facility because she still needed assistance with many activities of daily living, such as dressing, bathing, and transferring to and from bed. She had a dense left hemiparesis affecting both her arm and leg. She had received 1 month of standard speech therapy, 2 months of occupational therapy, and 6 months of physical therapy by the time she was brought to our facility. During preliminary testing for this study, participant 1 showed limited changes in rate, pause time, pitch, and loudness of speech. She did not have any signs of dysarthria but did have a mild left facial droop. She mentioned that she thought her speech was quieter and less expressive since her stroke.

Participant 2

Participant 2 was referred to the VA Brain Rehabilitation Research Center by an outpatient rehabilitation center. She lived at home with her spouse, who reported that she was dependent for most activities of daily living. She had left-sided weakness in both the upper and lower limbs. She had received 2 1/2 months of traditional speech therapy, 1 month of physical therapy, and 4 months of occupational therapy at the outpatient rehabilitation center before being referred to our center. Her caregiver reported that her voice conveyed less emotion than before her stroke. During our preliminary testing, participant 2 showed almost no changes in rate, pause time, pitch, or loudness in speech. She did not have any signs of dysarthria or oromotor or facial weakness and did not complain about her speech.

Participant 3

Participant 3 was referred to our center by a local rehabilitation hospital. He lived at home with family members. He had a dense left hemiparesis affecting both his upper and lower limbs. He had received 1 month of

Table 2.
Participant scores for batteries of emotional communication.

<table>
<thead>
<tr>
<th>Test</th>
<th>Participant 1</th>
<th></th>
<th>Participant 2</th>
<th></th>
<th>Participant 3</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Pre-Tx%</td>
<td>Post-Tx%</td>
<td>Pre-Tx%</td>
<td>Post-Tx%</td>
<td>Pre-Tx%</td>
<td>Post-Tx%</td>
</tr>
<tr>
<td>Florida Affect Battery</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Identity Discrimination</td>
<td>80</td>
<td>90</td>
<td>80</td>
<td>95</td>
<td>100</td>
<td>100</td>
</tr>
<tr>
<td>Affect Discrimination</td>
<td>90</td>
<td>75</td>
<td>90</td>
<td>80</td>
<td>70</td>
<td>75</td>
</tr>
<tr>
<td>Name Affect</td>
<td>80</td>
<td>100</td>
<td>85</td>
<td>75</td>
<td>75</td>
<td>90</td>
</tr>
<tr>
<td>Select Affect</td>
<td>100</td>
<td>100</td>
<td>90</td>
<td>85</td>
<td>90</td>
<td>95</td>
</tr>
<tr>
<td>Match Affect</td>
<td>85</td>
<td>85</td>
<td>75</td>
<td>80</td>
<td>75</td>
<td>85</td>
</tr>
<tr>
<td>Nonemotional Prosody Discrimination</td>
<td>81</td>
<td>93</td>
<td>81</td>
<td>75</td>
<td>81</td>
<td>100</td>
</tr>
<tr>
<td>Emotional Prosody Discrimination</td>
<td>95</td>
<td>100</td>
<td>100</td>
<td>100</td>
<td>90</td>
<td>95</td>
</tr>
<tr>
<td>Name Emotional Prosody</td>
<td>90</td>
<td>100</td>
<td>80</td>
<td>75</td>
<td>85</td>
<td>80</td>
</tr>
<tr>
<td>Conflicting Prosody</td>
<td>83</td>
<td>97</td>
<td>89</td>
<td>89</td>
<td>90</td>
<td>87</td>
</tr>
<tr>
<td>Match Emotional Prosody to Emotional Face</td>
<td>80</td>
<td>90</td>
<td>75</td>
<td>75</td>
<td>65</td>
<td>75</td>
</tr>
<tr>
<td>Match Emotional Face to Emotional Prosody</td>
<td>89</td>
<td>100</td>
<td>75</td>
<td>85</td>
<td>70</td>
<td>84</td>
</tr>
<tr>
<td>Expressive Battery</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nonemotional Prosody Imitation</td>
<td>80</td>
<td>100</td>
<td>100</td>
<td>100</td>
<td>75</td>
<td>100</td>
</tr>
<tr>
<td>Emotional Prosody Imitation (neutral semantic content)</td>
<td>40</td>
<td>70</td>
<td>65</td>
<td>100</td>
<td>35</td>
<td>65</td>
</tr>
<tr>
<td>Emotional Prosody Imitation (semantics congruent/incongruent)</td>
<td>30</td>
<td>65</td>
<td>65</td>
<td>95</td>
<td>20</td>
<td>60</td>
</tr>
<tr>
<td>Emotional Prosody to Command (semantics congruent/incongruent)</td>
<td>32</td>
<td>75</td>
<td>36</td>
<td>64</td>
<td>39</td>
<td>71</td>
</tr>
<tr>
<td>Emotional Prosody to Command (neutral semantic content)</td>
<td>35</td>
<td>70</td>
<td>40</td>
<td>70</td>
<td>30</td>
<td>65</td>
</tr>
<tr>
<td>Nonemotional Prosody to Command</td>
<td>100</td>
<td>90</td>
<td>65</td>
<td>75</td>
<td>95</td>
<td>100</td>
</tr>
</tbody>
</table>

Tx% = percentage correct either prior to (pre-) or following (post-) therapy
speech and occupational therapy and 2 months of physical therapy. His spouse reported that he was less animated now than before the stroke. She also reported that he seemed less concerned with her emotions. During preliminary testing for this study, participant 3 showed limited changes in rate, pause time, pitch, and loudness in speech. In addition, he had a mild dysarthria with consonant imprecision and a left facial droop. He also did not complain about his speech.

Treatment Procedures

This study investigated two mechanism-based treatments for expressive aprosodia, one an imitative treatment and the other a cognitive-linguistic treatment. Both treatments follow a six-step cueing continuum. Maximum cueing was provided in the first step and was systematically decreased as the participant progressed to the final step. For example, in the initial steps of the imitative treatment, the clinician provided a model sentence using the targeted emotional prosody and the participant attempted to imitate using the same emotional tone of voice. In the ensuing steps, the participant moved from imitation of clinician’s model to independent production of the sentence using the target emotional tone of voice. In the first step of the cognitive-linguistic therapy, the participant was provided cards listing an emotion name, the vocal characteristics of an emotional tone of voice, and a picture showing the appropriate facial expression for the target emotion. These cues were systematically removed as the participant successfully moved through each step. The steps for both experimental treatments and the stimuli used in each step are outlined in Appendixes A and B (available in the online version only).

Treatment Stimuli

We generated the treatment and outcome measure stimuli with the help of colleagues by compiling lists of sentences invoking an affective response. Of these sentences, those eliciting the strongest affective response for each emotion amongst our group of coauthors and colleagues were selected as stimuli. All sentences were semantically congruent with their accompanying emotional tone of voice. The sentences were divided into three main sets, one set to be treated during imitative treatment (20 sentences), one set to be treated during cognitive-linguistic treatment (20 sentences), and one set that was never treated but was used to sample generalization (30 sentences). In addition, a corpus of sentences was used for linguistic prosody (10 sentences). A representative sample of the stimuli sentences used in this study can be found in Appendix C (available in the online version only).

Experimental Design

A single-subject ABAC design with replication across three participants was employed. During the initial “A,” or no-treatment, phase, stable baselines for verbal production of five emotional tones of voice were established and verified via the “C” statistic [13]. Both treatment phases (“B” and “C”) were approximately 1 month in duration and consisted of 20 treatment sessions. The average treatment session length was 1 hour. Two treatments, one imitative and one cognitive-linguistic, were employed and treatment order was determined randomly. The second “A,” or no-treatment, phase was also 1 month in duration. Each treatment phase was immediately followed by two sessions of posttesting. One participant was randomly assigned to receive imitative treatment during the first treatment period, followed by cognitive-linguistic treatment during the second treatment period, and the other two were randomly assigned to the opposite order.

Outcome Measure

Treatment effect was measured by the administration, rating, and analysis of an outcome measure. This
measure consisted of sentences that the participant was asked to produce using targeted emotional tones of voice. The sentences targeted four treated emotions (happy, sad, angry, and neutral) and one control or nontreated emotion (fear). The corpus of stimuli included some sentences that were considered control items (both linguistic prosody and fearful tone of voice were never treated), some sentences that were considered generalization sentences (sentences that were never treated but were produced with the use of trained tones of voice—angry, sad, happy), and sentences that were actively treated.

The 50-item baseline outcome measure consisted of 10 linguistic prosody sentences, 5 fearful sentences, 10 sentences from the set used during imitative treatment, 10 sentences from the set used during cognitive-linguistic treatment, and 15 sentences from the set that was never treated but was used to sample generalization. The 45-item outcome measure administered prior to each treatment session was essentially the same, except that the number of items from the set that was currently being treated was dropped from 10 to 5. The sentences were rotationally ordered and balanced across emotions so that all sentences from all sets were probed equally over the course of treatment.

Outcome Measure Administration

The outcome measure was administered eight times during both pretreatment baseline phases (A phases), daily prior to the start of therapy (both B and C phases), and twice during the two posttesting sessions that directly followed both treatments. The entire measure consisted of 50 sentences for baseline phases and 45 sentences for therapy phases. We administered the outcome measure by presenting a sentence written on a card to the participant, who was then asked to say the sentence aloud using a particular tone of voice. For example, the participant was shown “Our house is on fire” and asked, “Please say this sentence using a fearful tone of voice.”

Outcome Measure Analysis

All outcome measures were audi-taped with a TASCAM digital audio tape recorder (model DA-P1, Montebello, CA). Each sentence was scored as “plus” if correctly conveying, or “minus” if incorrectly conveying, the requested emotional tone of voice. The therapist conducted the scoring online during the session, and this scoring was also later judged by a trained rater blind to the time of testing. The judgments of both the therapist and trained rater were based solely on verbal expression; facial expression was not a factor. The therapist was instructed to look away while the participant produced the sentence. This practice was explained to the participant in the first treatment session.

The trained rater was a speech-language pathologist with 2 years of experience in evaluating the prosody of emotionally intoned sentences. Training for this rater included familiarization with the descriptions of features for each emotion, including respective changes in pitch, loudness, and rate. The rater also took part in research group sessions in which tapes of aprosodic speakers were discussed and individual features rated. Acoustic analysis of selected pre- and posttreatment data subsequently supported the trained rater’s judgments. The trained rater’s judgments were the data used in all analyses.

Both intra- and interjudge reliability were calculated with the use of 20 percent of participant’s productions. Intrajudge reliability for the trained rater was acceptable (Kendall’s Tau of 0.75, p < 0.001). Interjudge reliability based on judgments by the trained rater and another experienced clinician (B.H.) was also acceptable (Kendall’s Tau of 0.79, p < 0.001).

The percentage of the participants’ correct productions of sentences using treated and untreated emotions were graphed for analysis. The linguistic prosody sentences were not included in the analysis at this time. The data were then analyzed visually and statistically.

Visual Analysis

Visual inspection of outcome measure data was completed by three judges, all speech-language pathologists, all with at least 3 years experience judging data via visual inspection. Each independently judged the stability of both baseline phases for each participant and then considered the relative slope and height of the data displays during the two treatment phases. The judges received similar directions for judging displays for the untreated emotion. The Figure displays the graphs the judges used for the visual analysis.

Statistical Analysis

For the statistical analyses, effect sizes [14] were calculated for each participant for each therapy. The effect size for the first treatment was calculated by subtraction of the mean of the correct responses on the 8 baseline outcome measures from the mean of the correct responses on the 20 therapy outcome measures, divided
Figure.
Percentage of correct productions of sentences from outcome measure with use of treated and untreated emotions by session: (a) participant 1, (b) participant 2, and (c) participant 3.
by the standard deviation (SD) of the baseline outcome measure data (M). We calculated the effect sizes for each participant for the second therapy in an identical manner using data from the second therapy. The formula to calculate the effect size (ES) is

$$ES = \frac{M_{\text{therapy}} - M_{\text{baselines}}}{SD_{\text{baselines}}}.$$ 

Cohen provides approximate ranges for effect sizes with a small effect being about 0.2 to 0.3, a moderate effect being about 0.5, and a large effect being about 0.8 to 1.0 [15].

### RESULTS

Three judges unanimously agreed that visual displays of outcome measure data (Figure) showed evidence of treatment effects from both treatments for all three participants. No evidence of generalization to the untreated emotion was noted for any participant. The results of the statistical analyses (Table 3) were consistent with the findings from the visual analyses. Examination of effect sizes confirmed modest to substantial treatment effects for both treatments in all three participants.

Participant 1 was 9 months poststroke and had a stable baseline performance. This participant showed a response to both treatments, although the first treatment, which was imitative, showed a relatively greater effect size. For participant 1, the effect sizes were 3.68 (large effect) for imitative and 2.76 (large effect) for cognitive-linguistic treatment. Baseline performance preceding the second therapy, cognitive-linguistic, was more variable than preceding the first, but still statistically stable. Treatment effects were maintained at posttesting.

Participant 2 was 6 months poststroke and had a stable baseline performance. For this participant the second treatment, which was imitative, showed a relatively greater effect size. Participant 2 showed an effect size of 0.660 (moderate effect) for cognitive-linguistic treatment and 2.54 (large effect) for imitative. Treatment effect for the first treatment, cognitive-linguistic, did not emerge until the final sessions. Following discontinuation of this treatment, performance seemed to return to pretreatment levels. Considerable variability occurred after introduction of the imitative treatment, but the overall treatment effect was higher than during the cognitive-linguistic treatment. As with participant 1, a treatment effect was maintained at posttesting following the second treatment.

After a stable baseline period, participant 3, who was 4 months poststroke, showed an active response to both treatments. The treatment effect for the first treatment, cognitive-linguistic, was substantial. Participant 3 showed a treatment effect of 11.51 (large effect) for the cognitive-linguistic treatment and 2.01 (large effect) for the imitative treatment. Treatment effect for the first treatment, cognitive-linguistic did not emerge until the final sessions. Following discontinuation of this treatment, performance seemed to return to pretreatment levels. Considerable variability occurred after introduction of the imitative treatment, but the overall treatment effect was higher than during the cognitive-linguistic treatment. As with participant 1, a treatment effect was maintained at posttesting following the second treatment.

### DISCUSSION

The current study is a phase I investigation of mechanism-based treatments for expressive aprosodia. The data from these three participants provide evidence to support our hypothesis that both treatments are active. The effect sizes for the two treatments are moderate to large by traditional standards [15] and are within the range of effect sizes in the aphasiology literature for a variety of treatments and aphasia diagnoses [16–17].

Participants 1 and 2 showed greater treatment effects for the imitative treatment, and participant 3 for the cognitive-linguistic treatment. In a previous study, all participants were randomly assigned to begin with cognitive-linguistic treatment followed by imitation [9]. Since all had

<table>
<thead>
<tr>
<th>Participant</th>
<th>Treatment</th>
<th>Baseline Mean</th>
<th>Baseline SD</th>
<th>Therapy Mean</th>
<th>Therapy Z-Score</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Imitative</td>
<td>16.14</td>
<td>42.37</td>
<td>4.06</td>
<td>8.79</td>
</tr>
<tr>
<td>2</td>
<td>Cognitive-Linguistic</td>
<td>23.62</td>
<td>28.5</td>
<td>4.50</td>
<td>4.07</td>
</tr>
<tr>
<td>3</td>
<td>Cognitive-Linguistic</td>
<td>30.5</td>
<td>81.35</td>
<td>3.58</td>
<td>6.11</td>
</tr>
</tbody>
</table>

Table 3. Effect sizes (Z-scores) for change associated with first and second therapies (SD = standard deviation).
received the same treatment order, separating treatment effects from order effects was impossible. In the present study, participant 1 received imitative first and its treatment effect was larger than that of the cognitive-linguistic treatment. This participant’s response demonstrates that the imitative treatment’s activity does not depend on being preceded by cognitive-linguistic treatment.

It is noteworthy that generalization to the untreated emotion (fear) did not occur for any of our participants. However, data from the unpublished expressive battery suggest that all participants did generalize the treated emotions to untreated sentences. All participants showed improvement on both the imitative and to-command emotional prosody subtests of the expressive battery. However, because this tool is still undergoing standardization, the data from the expressive battery will not be analyzed statistically until a larger sample has been treated. Receptive aprosodia was also assessed both before and after treatment, and although the study focused on treating expressive aprosodia, all participants showed improvement on the receptive measure as well.

CONCLUSIONS

We hypothesized that both the imitative treatment and the cognitive-linguistic treatment would be active, but that the imitative treatment would show greater relative activity because of the assumption that a primarily expressive aprosodia is induced by a programming/planning deficit [9]. Also, the imitative treatment relies heavily on the participant’s ability to aurally discern emotional prosodic contours in order to imitate them accurately taking advantage of these participant’s relatively stronger receptive emotional communication skills. Participants 1 and 2 both showed the greatest effect sizes for the imitative treatment, as predicted. In contrast, participant 3 showed the greatest effect for the cognitive-linguistic treatment, which was the first treatment received. However, the second treatment, the imitation treatment, was limited by a ceiling effect.

Despite the limitations inherent in a phase I study, the evidence it provides of treatment effects for aprosodia is important. Normal human relationships are handicapped by an inability to signal emotion, which has functional consequences for those persons affected, as well as their family and friends. Therefore, active treatments for aprosodia raise the possibility of attaining a functional impact on this deficit. The three participants in this investigation manifested differing degrees of anosognosia clinically, but all three were faithful in their attendance of treatment sessions, and all three improved. This suggests that aprosodia may be amenable to behavioral treatments.

Further study of the treatment of expressive aprosodia is needed and more participants are currently being treated. Additional questions should be addressed, including the relative activity of the cognitive-linguistic versus the imitative treatments, the relationship of each treatment to receptive deficits, the influence of lesion site and size on treatment activity, and the ecological implications of the two treatments.

REFERENCES


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