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Imitation-based aphasia therapy increases narrative content: A case series

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Abstract

Objective—To test the generalization of an intensive imitation-based aphasia therapy to an unrelated narrative production task.

Design—ABA design study (A= no treatment; B= treatment) comparing imitation therapy to a baseline condition (pre-therapy). Participants produced narratives at two pre-therapy and two post-therapy time points. Narratives were analyzed for correct information units to determine the number and percent of communicative words produced.

Setting—A university laboratory and participants' homes.

Participants—Nineteen people with chronic aphasia following left hemisphere stroke.

Interventions—Six weeks of intensive imitation therapy $(3 \times 30 \text{ minutes/day}; 6 \text{ days/week})$ of words and phrases delivered via dedicated laptop.

Main measures—We performed t-tests to assess post-therapy changes in narrative production, as well as for intervals during which no intervention was provided. We used stepwise regression to examine the predictive value of demographic, behavioral, and neurological variables in determining treatment outcome.

Results—Significant gains were made on the narrative production task in both the number (mean = 34.36; p = 0.009) and percent (mean = 3.99; p = 0.023) of correct information units produced. For percent of correct information units, the number of therapy sessions completed was the sole predictor of changes in production following therapy (r= +0.542; p = 0.020). No variables predicted change in number of correct information units produced. There were no significant differences between the two pre-therapy or the two post-therapy time points (p > 0.294).

COMPETING INTERESTS

The Authors declare that there are no competing interests.

AUTHOR CONTRIBUTIONS

ESD conducted the analysis and wrote a full draft of the manuscript. SLS envisioned the study, mentored the first author, and edited the manuscript.

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Conclusions—Intensive imitation-based aphasia therapy may promote generalization to an unrelated narrative production task. Further investigation is indicated.

Keywords

aphasia; generalization; neurological rehabilitation; speech-language therapy; stroke

INTRODUCTION

Aphasia is defined by acquired impairments in language and communication skills that have significant ramifications for social and vocational experiences and quality of life. The broad impact of the disorder speaks to the need for achieving therapeutic effects across a variety of situations and communicative demands. However, failure to achieve generalization with aphasia therapy has been demonstrated across many measures, including naming (1), discourse comprehension (2), and the production of scripts (3). As systematic review indicates that all aphasia therapies are grossly equivalent in their outcomes, and limited in their direct effects (4), new therapeutic avenues must be explored (5).

Imitation-based aphasia therapy is a type of *action observation therapy* (6), in which engagement of the shared anatomical network underlying action observation and execution can enhance function after neurological injury. In our aphasia rehabilitation, we aimed for the observation and execution of speech (7), using observation to enhance the ability of individuals with aphasia to produce verbal output (8). We have previously shown this treatment to have benefit for the practiced task (i.e., repetition of words and phrases) as well as additional effects on naming and word finding tasks (9) from the Western Aphasia Battery (10). These past findings suggest that improvement following imitation-based treatment extends beyond trained items and is not restricted to the practiced task.

The current investigation thus hypothesizes that the imitation-based therapy generalizes to narrative production. In particular, we examine the changes of our patients on the "Cinderella" task (11), i.e., a task that assesses their ability to produce this well-known fairytale. We further seek to understand the relationship of select demographic, behavioral, and neurological variables with this benefit.

METHODS

Participants

Twenty-two native English speakers with chronic aphasia following left hemisphere ischemic stroke, confirmed by neurological examination and magnetic resonance imaging, were recruited for the study. Participants were recruited from individuals referred to the Rehabilitation Institute of Chicago for aphasia therapy. Participants were required to have sustained a single unilateral left hemisphere stroke resulting in aphasia > 5 months prior to study enrollment, and to have adequate vision, hearing, and cognitive abilities to participate in the therapy. Of the 22 individuals originally recruited, three decided not to participate prior to the initiation of therapy, leaving 19 experimental participants. The Western Aphasia Battery-Revised (WAB-R) was administered to obtain aphasia classifications and severity

ratings (Aphasia Quotient). Select demographic and neurological information for each of the 19 participants can be found in Table 1. Supplementary Figure 1 also identifies individual participants by their aphasia classification.

This study was approved by the Institutional Review Boards of The University of Chicago and the University of California, Irvine. Consent was obtained according to the Declaration of Helsinki.

Communication Outcome Measures

At each of four time points (Week –6: 6 weeks prior to therapy; Week 0: immediately prior to therapy; Week 6: immediately post-therapy; Week 12: 6 weeks post-therapy; see Supplementary Figure 2 for design), narratives were elicited by having participants recount the Cinderella fairytale aloud (11). These spoken narratives were recorded, transcribed, and subsequently analyzed for the number of correct information units (12) by a speechlanguage pathologist who was uninvolved in the therapy and blinded to the time points at which the narratives were collected. Words were scored as correct information units if they were intelligible, novel during the retelling of the story, and relevant to the story. The percent of correct information units was also calculated by dividing the number of correct information units by the total number of words produced at each time point, as this is a measure that reliably separates individuals with aphasia from healthy controls (12). Individuals with aphasia not only communicate less information (resulting in lower number of correct information units), they also have more unsuccessful attempts to communicate information, such as producing an inappropriate word or repeating the same word many times (resulting in lower percent of correct information units). This ratio of communication successes to total communication attempts, which could be conceived of as the efficiency of information transfer, is captured in the percent of correct information units produced. These changes in percent are absolute (i.e., changes in the proportion of words produced that were accurate and informative), rather than relative (i.e., a percentage change from an individual's baseline performance).

Study Design

Behavioral evaluations were performed at two separate time points before therapy (baseline assessment) and two time points after therapy. Baseline evaluations occurred 6 weeks before therapy (Week –6) and then at the onset of therapy (Week 0). Post-therapy evaluations occurred immediately following six weeks of therapy (Week 6) and then 6 weeks after that (Week 12). The study design is depicted in Supplementary Figure 2.

Therapy

Participants completed 6 weeks of intensive imitation-based therapy (three 30 minute sessions, 6 days per week) administered via a preprogrammed, dedicated laptop computer. A trained speech-language pathologist administered one session per week in person at the Rehabilitation Institute of Chicago, and the others were performed at home, using the borrowed laptop computer. The therapy required participants to listen to words and phrases presented by six different speakers ("talkers") and to repeat them orally six times. Stimuli were selected to be high frequency and ecologically valid (i.e., real words and phrases

that might be used by an English speaker in the course of daily activities). Words and phrases used as stimuli were unrelated to the Cinderella fairytale, which was used as the communication outcome measure in this generalization study. A trained speech-language pathologist adjusted stimulus complexity (length of words and phrases as well as phonetic features, such as consonant clusters) on an individual basis based on pre-therapy repetition ability. With informed consent, we monitored (and encouraged) compliance during home therapy sessions by recording all sessions via the camera and microphone of the laptop computer. Additional information about action observation therapy in general (13) or this particular imitation-based aphasia therapy (8, 9) are available in the literature and in Supplementary Material.

Statistical Analysis

One-tailed paired t-tests ($\alpha = 0.05$) were used to compare number and percent of correct information units produced pre-therapy (mean of the two baseline scores) vs. immediately post-therapy (Week 6) due to our strong *a priori* hypothesis that the therapy would increase productive output. Two-tailed paired t-tests were used to compare the number and percent of correct information units produced between the two baseline sessions, and between the two post-therapy sessions. One participant (#20) was excluded due to missing data, leaving 18 participants. An additional participant (#10) missed the Week 12 assessment and was excluded from the t-test comparing the two post-therapy sessions, leaving 17 participants for this comparison.

Two separate stepwise regression analyses were used to identify variables associated with changes in number of correct information units and percent of correct information units produced following treatment (Week 6 compared to mean baseline performance). Regression was performed using the MATLAB stepwise function with the default settings using a criterion of $\alpha=0.05$ to select new predictors and $\alpha=0.10$ to exclude existing predictors. Variables included as potential predictors in the regression model included age, months post stroke (MPS), number of therapy sessions completed (NTS), fluency (fluent vs. nonfluent), lesion size, and baseline performance (i.e., mean number or percent of correct information units produced before the onset of therapy). Values for these variables are shown in Tables 1 and 3, with the exception of fluency, which was determined based on aphasia classification. Participants with Broca's (n=9) and transcortical motor (n=1) aphasia are nonfluent. Participants with anomic (n=6), conduction (n=1), and Wernicke's (n=1) aphasia are fluent. More precise aphasia classifications were not used in the regression due to limited representation of several categories.

A Pearson correlation coefficient was calculated for any variable found to be significantly predictive of either outcome (i.e., change in number or percent correct information units) with the predicted outcome measure.

RESULTS

Participants ranged in age from 31 to 72 years (mean = 54; SD = 11.37; 4 female (21%)). All participants sustained a single stroke 5 to 130 months prior to enrollment (mean = 41.63; SD = 42.94). Aphasia Quotient on the WAB-R ranged from 20.50 to 93.15 (mean = 67.64;

SD = 20.00). Lesion size ranged from 14.11 to 219.06 cubic centimeters (mean = 89.41; SD = 52.37).

Mean scores for the group (both number and percent of correct information units) are shown in Table 2 for each of the four time points. For each participant, pre-therapy (the average of the two pre-therapy scores) and post-therapy (Week 6) scores (both number and percent of correct information units) are shown in Table 3. Individual compliance (number of sessions completed) is also shown in Table 3.

The mean change in number of correct information units produced for the narrative task from pre- to post-therapy (Week 6) was 34.36 (SD = 55.15; range -58 to 129). This increase was significant at $\alpha = 0.05$ (t(17) = 2.64; p = 0.009) with an effect size of 0.377. There were no significant differences between the two pre-therapy sessions (t(17) = 0.17; p = 0.864) or the two post-therapy sessions (t(16) = 0.50; p = 0.622). Individual performance for each of the four time points is shown in Supplementary Figure 3.

The mean change in percent of correct information units produced for the narrative task from pre- to post-therapy (Week 6) was 3.99 (SD = 7.88; range -15.01 to 15.20). This increase was significant at $\alpha = 0.05$ (t(17) = 2.15; p = 0.023) with an effect size of 0.215. There were no significant differences between the two pre-therapy sessions (t(17) = 0.38; p = 0.712) or the two post-therapy sessions (t(16) = 1.08; p = 0.294). Individual performance for each of the four time points is shown in Supplementary Figure 4.

Stepwise linear regression performed to identify variables significantly predictive of the change in number of correct information units produced following therapy selected no variables. For the change in percent of correct information units produced following therapy, the sole variable selected was the number of sessions completed ($R^2 = 0.29$; F(1,17) = 6.67; p = 0.020).

This variable was found to have a significant positive correlation with post-therapy change in percent of correct information units produced during the narrative task (r= +0.542; p = 0.020). Supplementary Figure 5 shows the relationship between these variables.

DISCUSSION

We found that patients with aphasia demonstrate significant improvement in narrative ability following a six-week period of intensive therapy involving repetition of unrelated words and phrases. The participants in this study significantly increased both the number and percent of correct information units that they produced following the course of therapy. There were no significant differences in either of these measures during the six weeks before or after the therapy course. Additionally, we found that the number of sessions completed was predictive of improvement for percent, but not number, of correct information units.

These behavioral outcomes are of particular interest as the practiced task (repetition) is quite dissimilar to the measure on which improvement was demonstrated (narration). Failure to achieve generalization is a significant obstacle in aphasia treatment, and the most common scenario is for therapeutic gains to be restricted to precisely the task(s) and items that are

explicitly trained, with no effect on untrained items (14). However, recent studies exploring speech entrainment, or online imitation, have found generalization to untrained scripts as well as to spontaneous speech (15). These findings, in combination with ours, may suggest a unique benefit of imitation-based therapy.

The changes found here indicate that individuals with aphasia are able to produce narratives that are more informative (i.e., larger number of correct information units) and efficient (i.e., higher percentage of correct information units) following intensive imitation practice, despite the fact that neither the narrative task nor the specific words and phrases used in the task are related to the content of training. Additionally, the lack of significant differences between the end of treatment and six weeks later suggests that our participants were able to maintain these benefits once achieved. This is clinically relevant as most studies – even with large therapeutic benefits – do not show maintenance of therapeutic benefits into the period following cessation of treatment (e.g., (15, 16)).

As with any intervention, individual compliance varied in our sample, and some participants completed far fewer sessions than others. We found a positive correlation between change in percent of correct information units produced and the number of sessions completed over the course of treatment. This is perhaps unsurprising, as more treatment (17) and higher intensity (18) are associated with better outcomes following therapy. However, it may be unexpected that this measure trumps other variables previously found to be associated with aphasia prognostication, such as time post stroke (19) and lesion size (20).

We designed the imitation-based therapy to incorporate high intensity. It is theorized that the sort of massed stimulation provided by our treatment program supports implicit learning through the repeated engagement of neural pathways that are consequently strengthened and/or formation of new pathways (21). While the present study does not explore the brain changes underlying our findings (but see (22)), generalization following aphasia therapy, such as that demonstrated by our participants, is associated with changes in functional connectivity as measured by functional magnetic resonance imaging (23). Further suggestion that synaptic changes facilitate generalization can be found in the literature on transcranial direct current stimulation, a form of noninvasive brain stimulation that promotes generalization, as well as maintenance, when paired with aphasia therapy (24, 25).

There is strong evidence for causality in the relationship between our therapy and changes on the narrative task, as significant changes occur only over the duration of therapy. Despite notable intraindividual variability, no significant changes occur during the two equally spaced pre-therapy assessments, or during two similar post-therapy sessions, providing confidence that the therapy itself produced the observed effect. Still, the significant positive correlation between the number of sessions completed and the change in percent correct information units produced may or may not be causal. It is possible that participants who did not feel that the therapy was benefiting them were less motivated to complete as many sessions, and that poor performance caused fewer sessions to be completed. We do not subscribe to this conclusion, however, as individuals with poor compliance completed fewer sessions consistently throughout the entire 6-week duration of treatment, and did not reduce participation over the course of the program. Thus, we do not believe that these participants

demonstrated waning enthusiasm for the therapy. However, it remains possible that another factor such as attention – known to be impaired in aphasia (26) – may have resulted in both limited participation and lesser benefit of therapy.

While we believe the current study offers provocative findings, there are several limitations. First, the sample size, while reasonable for a study of this type, is small, reducing power. This may have contributed to our failure to identify a predictor for changes in the number of correct information units produced. Second, an unexamined variable, such as attention, may have impacted both therapy compliance and improved performance following therapy. It would be wise to include an attentional control in similar future studies. Further, performance at the two baseline time points was quite variable despite the lack of statistically significant differences. Including additional pre-therapy time points could have established a more stable baseline and better controlled for possible practice effects. Finally, there was a great deal of variability in individual changes following therapy, and it therefore remains unclear which individuals might best benefit from this type of intervention.

The present treatment study uses a biologically motivated approach to aphasia therapy (27, 28) and finds significant generalization beyond the practiced task. Furthermore, the more therapy sessions completed, the greater the therapeutic benefit. This is interpreted as support for our hypothesis that by engaging populations of neurons active during the observation and execution of speech, we are strengthening those networks' ability to support the production of speech. By using a general approach that targets the processes subserving imitation, we are able to achieve generalized improvement in the domains of speech and language. While further studies are needed due to the considerable variability in individual outcomes, such action observation therapy may ultimately allow our patients to carry greater benefits out of our clinics and into their daily lives.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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CLINICAL MESSAGES

• Imitation of words and phrases facilitates generalization to narrative production in aphasia

• Intensity of therapy correlates with degree of benefit

Table 1

Select demographic and neurological information for the 19 therapy participants. This table includes sex, age, months post stroke (MPS), aphasia quotient (mean of two pre-therapy time points) and aphasia classification (both from the Western Aphasia Battery-Revised), and lesion size (in cubic centimeters), as well as mean values and standard deviations for numerical data.

1 F F M A A M M A A M M A A M M A A M M A A M					()
	72	11	30.40	Broca's	61.88
	09	2	08.99	Broca's	50.46
	63	L	20.50	Broca's	45.38
	99	16	78.05	Broca's	49.21
	99	8	79.25	Conduction	42.12
	46	28	76.35	Broca's	130.58
	31	11	79.75	Anomic	158.29
	28	13	57.90	Trans. Motor	137.13
	55	22	91.20	Anomic	14.11
13 M	36	82	77.65	Broca's	86.93
14 M	37	51	66.35	Broca's	69.06
15 M	70	120	82.80	Anomic	219.06
16 M	58	29	93.15	Anomic	48.66
17 M	57	130	78.15	Anomic	82.35
18 M	55	81	53.80	Wernicke's	162.10
19 M	42	124	83.25	Anomic	36.58
20 M	09	L	63.70	Trans. Sensory	100.62
21 M	43	15	37.20	Broca's	81.98
22 M	49	29	69.10	Broca's	100.62
Mean (SD)	54 (11.37)	41.63 (42.94)	67.65 (20.00)	-	89.41 (52.37)

Table 2

Mean group performance at each time point. Mean (standard deviation) for number and percent correct information units (CIUs) produced pre-therapy (weeks -6.0) and post-therapy (weeks 6.12). Percent of correct information units are calculated by dividing the number of correct information units by the number of total words produced, and thus reflect the ratio of successful communication to total communication attempts.

CIUs	Week -6	Week 0	Week 6	Week 12			
Number	72.33 (87.41)	67.21 (83.02)	105.94 (116.25)	96.88 (127.45)			
Percent	31.27 (24.75)	30.52 (24.52)	35.64 (26.53)	32.43 (23.76)			

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Table 3

Individual compliance and pre-/post-therapy performance on the Cinderella narrative task. Pre-therapy scores are individual means from the two baseline sessions (Weeks -6, 0). Week 6 scores were obtained immediately following therapy.

Week 6 CIUs (%)	0	0	0	38.3	43.8	48.5	42.5	20.4	8.07	27.7	30.0	65.2	70.0	67.4	1.1	0.09		10.3	15.7	
Pre-therapy CIUs (%)	0	15.0	0	30.0	38.6	40.7	43.0	5.2	70.4	49.9	18.0	61.4	56.1	54.0	9.0	63.2	3.2	7.3	7.9	
Week 6 CIUs (#)	0	0	0	119	244	33	178	10	160	45	32	200	270	190	3	383		14	26	
Pre-therapy CIUs (#)	0.0	10.0	0.0	30.5	146.5	22.5	92.0	2.5	145.0	41.5	30.0	91.5	146.5	248.0	16.5	254.0	2.0	7.0	4.5	
NTS	88	84	54	06	06	108	106	101	108	108	105	66	108	107	79	53	103	108	108	
Participant	1	2	4	5	9	6	10	11	12	13	14	15	16	17	18	19	20	21	22	

Abbreviations are as follows: NTS: number of therapy sessions; CIUs: correct information units; #: number; %: percent.