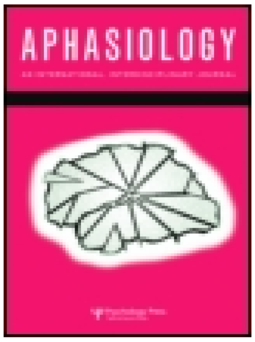

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Intensive sound production treatment for severe, chronic apraxia of speech

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ABSTRACT

Background: Massed practice has become a widely used method of treatment administration in the study of aphasia. It is often shown to be as or more effective than treatment provided in a distributed fashion. Some studies report changes beyond the targeted treatment materials, and durability of effects tends to be positive. Feasibility of this specific dosage for Sound Production Training (SPT) for apraxia of speech has yet to be tested.

Aims: This case study investigates the effects of a massed practice administration of SPT for a patient with severe and chronic apraxia of speech. Responsiveness was monitored for target treatment items at the level of the word and the manner of articulation. In addition, we assessed for changes in general aphasia severity, patient perception of communicative effectiveness, and ability to self-correct errors.

Methods & Procedures: SPT was administered for 3 h per week-day over 2 weeks. All sessions were audiotaped and analyzed independently by two raters. Initial responses were coded as correct or incorrect and then re-analyzed for each manner of articulation within each word. The total number of correct and incorrect self-corrections were also recorded. Multiple baselines across behaviors was used to track progress on trained materials.

Outcomes & Results: Improvements were observed on the accuracy of trained repeated productions, untrained equivalent exemplars, increased self-corrections, and also in reduced aphasia severity. There was no observed benefit to increased repetitions of a specific manner of articulation.

Conclusions: Massed practice is a viable way to administer SPT and may also positively impact non-targeted treatment items and objectives. Self-correction attempts precede or coincide with production accuracy and may be worth training in patients who do not spontaneously self-correct. Since increased repetitions did not improve a specific manner of speech, we speculate that the use of functional words versus those targeting specific phonemes may be as effective at eliciting desired speech outcomes.

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Apraxia of speech; intensive treatment; massed practice; sound production treatment; neuroplasticity

Apraxia of Speech (AOS), a motor speech disorder that often accompanies an aphasia diagnosis post cerebral vascular accident (CVA), refers to impairment in the planning and programming of speech. It may be difficult to assess based on common co-

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occurrence with aphasia and therefore evidence for treatment remains limited (Ballard et al., 2015). Since 2006, there has been some increase in replication studies for AOS, but more are needed (Ballard et al., 2015). A number of treatment approaches have been developed targeting this disorder but there are few positive reports of generalization following them (Ballard, 2001). Limited generalization is also an important consideration within the aphasia literature but has become a greater area of focus in light of recent attention on intensity of treatment. Evidence from the Cochrane review (Brady, Kelly, Godwin, Enderby, & Campbell, 2016) points to better functional communication outcomes for those who received more speech and language therapy than less but also note a higher dropout rate. Limited standardization of dosage parameters combined with the unknown contribution of other important treatment variables make it difficult to interpret the large body of aphasia literature at this time (Raymer et al., 2008). It seems it would be more straightforward to examine the application of intensive treatment to AOS in which treatment tends to have less variation and the parameters tend to be clearly defined.

Sound Production Treatment (SPT) is one type of treatment of AOS. It was developed to improve the articulation of difficult-to-produce sounds (Wambaugh, West, & Doyle, 1998) and may be the most extensively studied treatment for apraxia of speech (AOS) to date. Consistently positive changes in articulation have been documented for treated sounds with limited generalization observed for untreated sounds. Most studies using SPT have been based on individuals with moderate-severe AOS but consistent effects were reported for one participant with severe AOS and aphasia by Wambaugh and Mauszycki (2010) and and for one of the five participants in a recent study by Wambaugh et al. (2018). Phase II efficacy studies (Robey, 2004) have begun with initiation of investigation on the dose frequency and schedule of treatment sessions (Wambaugh, Nessler, Cameron, & Mauszycki, 2013; Wambaugh et al., 2018). The current investigation, which used a single-subject experimental design to document behavioral changes using an intensive dosage to treat severe AOS, contributes to this effort.

In the current study, we applied SPT methods described by Wambaugh et al. (2013); however, treatment was provided at 30 h over 2 weeks. This massed practice schedule is reported to have positive effects in multiple aphasia treatment studies (Johnson et al., 2013; Maher et al., 2006; Meinzer et al., 2004; Mozeiko, Coelho, & Myers, 2016; Pulvermuller et al., 2001) but this particular dosage schedule has yet to be used in studies of AOS treatment. Here, we use Warren, Fey, and Yoder (2007) operational definitions when describing dosage parameters. In the current study, the dose frequency was comparable to that administered by Wambaugh et al. (2013) at 3 h per day for 5 days per week (15 h total) but we increased the total intervention duration to 2 weeks for a total of 30 h of SPT.

Generalization beyond trained stimuli appears to be the case in the single-subject data presented in some studies that have targeted oral-verbal language using this dosage schedule. In them, it is not uncommon to observe generalization to untreated modalities following treatments with relatively high session durations (hours long) and relatively short total treatment durations (2 weeks). Specifically, these have included improvements in comprehension (Kirmess & Maher, 2010; Richter, Miltner, & Straube, 2008; Szaflarski et al., 2008) in semantic knowledge (Richter et al., 2008), gains in reading and writing (Kirmess & Maher, 2010; Mozeiko, Myers, & Coelho, 2014, 2018) and also on cognitive measures (Mozeiko et al., 2014b). Given the close proximity and interconnectedness of speech and language brain regions, we predicted that intensive training and resulting improvements in speech production would have an effect on overall aphasia severity well.

Determining the feasibility of administering SPT with a very rigorous schedule and measuring treatment responsiveness was a primary objective in this study. In addition, we aimed to provide detailed data on the quality of verbal productions and the participant's ability to self-correct over the course of treatment. These data not only provide insight into the course of recovery for an individual with severe AOS, but may also influence clinicians' approach to remediation and documenting change. To this end, we performed both an error analysis and a characterization of the participant's ability to self-correct his errors. Counting the number of errors per manner (e.g., affricates, stops, fricatives, etc.) was a means of judging consistency of productions. Inconsistency of errors has long been a hallmark of AOS diagnosis (http://www.asha.org/public/speech/disorders/ApraxiaAdults/) however, some evidence points to more consistent patterns (Haley & Martin, 2011; Mauszycki, Wambaugh, & Cameron, 2010). Understanding these patterns is necessary in order to differentially diagnose AOS as distinct from aphasia and is important in providing treatment for AOS. Mauszycki and colleagues demonstrated that error patterns, particularly distortions, tended to persist and that different error types, such as substitutions, were infrequently produced (Mauszycki et al., 2010), consistent with previous findings by McNeil and colleagues (McNeil, O'Dell, Miller, & Hunter, 1995). In addition, we investigated improvements over time within each manner of articulation to assess whether some manners were more responsive to this intensive treatment or whether changes were consistent across manners of articulation. We hypothesized that manners of articulation that were repeated more frequently would improve more than those manners of articulation that were less practiced.

Important to determining potential responsiveness to treatment is the ability to self-monitor. Identifying errors by attempted self-correction is thought to be a natural precursor to improvement (Rosenbek, Lemme, Ahern, Harris, & Wertz, 1973). Van der Merwe and colleagues reported increased accurate self-corrections as a result of treatment and decreased self-corrections overall but the percentage of attempted self-corrections on incorrect productions remained stable, which authors interpreted as a lack of improvement in processes of planning and programming of speech, referred to as feedforward motor control by Van der Merwe et al. (2007). In the current study, attempted and accurate self-corrections were monitored and results are compared to findings by Van der Merwe (2007).

To summarize, in this study methodical analysis of treatment response of a single individual with severe AOS was performed in order to:

(1) Assess responsiveness to an intensive dosage schedule (30 h over 2 weeks) of SPT for someone with severe and chronic AOS.
(2) Determine whether participating in SPT also decreases aphasia severity as determined by the Western Aphasia Battery Aphasia Quotient (WAB-R AQ; Kertesz, 2006) and by results of the Communication Effectiveness Index (CETI; Lomas et al., 1989)

(3) Investigate whether more repetitions in a specific manner of articulation results in more consistent productions, more complete mastery, and better maintenance than manners that received fewer repetitions.
(4) Examine the role of self-monitoring in treatment as related to production accuracy.

Methods

Participant

The participant, "Mark" was a 51-year-old male with mild oral, non-speech apraxia according to the Apraxia Battery for Adults (ABA-2) (Dabul and Dabul, 2000) and chronic, severe verbal AOS characterized by groping, distorted substitutions and delayed or absent response initiation and moderate-severe Broca's aphasia (WAB-R AQ – 27.4) following a single stroke. At the start of the study, Mark's spontaneous productions were limited to the word "no" and to one expletive. When asked to repeat items during the baseline testing he was asked repeatedly to take larger breaths in order to be heard. Pre-treatment, his voice quality was hoarse, breathy, and generally weak. Mark had been a Naval officer, was right handed, a native English speaker with no reported history of language disorder, substance abuse, or neurological or psychological conditions. He passed a pure-tone hearing screening at 35 dB in each ear. At the time of the investigation, he was 3.6 years post-hemorrhagic CVA which resulted in a large infarct documented by the hospital radiologist as "extending to subcortical regions impacting the left thalamus and basal ganglia." Mark had a significant right-sided hemiparesis at the time of the study and had been discharged from speech language therapy due to performance plateau. He did not participate in any other speech or language interventions during the period of investigation. Mark did not receive compensation for participation in this investigation (approved by University IRB).

Stimuli

Twelve sounds were selected for treatment based on pre-treatment assessment. Wambaugh's SPT-Supplemental Materials (Wambaugh, n.d.) were used to determine production accuracy of monosyllabic stimuli. According to Wambaugh, lists should be comprised of real words and with a variety of vowels. She suggests that 1–3 sounds are treated at one time. Neighborhood density is not a suggested consideration during list creation but Wambaugh does suggest creating lists that include higher frequency items when possible and using words in which errors result in homonymy.

Using these guidelines, 12 monosyllabic words that had contrastive minimal pairs were created using each sound: eight trained, four untrained. Sounds were grouped into four-word lists containing three sounds each. Each list had a total of 24 trained and 12 untrained words: List 1-/initial g, s, l/; List 2-/initial k, f, r/; List 3-/initial θ, d͡ʒ, and initial l-blends/; List 4-/initial s-blends, v, and final k/(see Table 1).

When possible, minimal pairs were used with contrastive target sounds that the client was able to repeat with some consistency during baseline testing. For example,/b/ and

Table 1. Experimental Stimuli.

List 1-blocked	List 2-random	List 3- blocked	List 4-random
	Trained Items		
guy	core	thin	ski
gay	cap	thumb	star
gore	Kay	Thor	sled
got	cot	thought	swing
game	came	thaw	small
gill	keep	thick	spin
go	Ken	thank	stem
gas	kin	thief	skit
say	fin	clown	vine
sit	fame	black	voom
sill	fear	flag	veer
sue	fit	sled	vet
Sam	face	glass	vase
seal	feet	plow	via
sew	fell	blue	van
see	fan	glue	vee
lie	wren	joke	puck
low	right	Jane	gawk
lit	rail	Jill	sack
Lee	rash	juice	tack
lay	rack	jet	Rick
lick	rip	jab	tick
line	rat	join	pack
late	row	job	tuck
	Untrained Items		
gun	comb	thigh	step
gal	cut	thorn	slim
get	cat	thing	spill
gate	kit	third	skin
sip	fat	clan	vat
soon	fed	slide	vex
set	fun	play	vie
sat	fill	bled	vim
lip	ripe	jam	duck
load	ram	gem	deck
light	rim	June	pick
lame	wreck	jut	book

/p/ were produced with some reliability so minimal pair choices for /guy/ was /buy/ and for /gore/ was /pour/.

Design

This study was originally conceptualized as a single case experimental multiple baseline across behaviors in order to track accuracy of trained and untrained items. We did not include a sufficiently different untrained list, however, to demonstrate that the training is responsible for any potential changes. Our untrained list was designed to mirror the trained list and is a replication of the types of untrained lists provided in studies by Wambaugh; however, Wambaugh et al. (2013) make use of a reduced probing schedule to avoid a practice effect. Since we did not reduce the probing schedule, our untrained items cannot be considered a true control probe. Therefore, we now present this data as a case study.

Correct articulation of the full word was necessary in order to be scored as a correct production during probes. When 80% accuracy was achieved in three consecutive probes, the next list was introduced. A nonrising baseline was established after five probes for all lists prior to treatment. Treatment began with the first list of 24 words and baselining of the yet-to-be-trained second list continued. Mastery of a list was determined when 80% accuracy was achieved in three consecutive trials at which time the second list was initiated, and baselining of the third list began. Probes continued after criterion was met on each list in order to assess maintenance.

Probes were conducted prior to each of the 3 h of daily treatment during which no feedback was provided in regard to articulatory accuracy. Follow-up probes were conducted 1, 4, and 10 weeks post the final treatment session, as was the WAB-R AQ to gauge aphasia severity and the Apraxia Battery for Adults (ABA) to assess potential change in severity of AOS.

Treatment

The SPT protocol outlined in Wambaugh and colleagues' multiple investigations (i.e., Wambaugh et al., 2014; Wambaugh & Mauszycki, 2010; Wambaugh et al., 2013, 1998) and in *SPT: General Instructions for Clinical Application* (Wambaugh, n.d.) was replicated closely. In the first step of this response-contingent treatment, the SLP says the word and requests a repetition of the target word. If correct, the participant is asked to repeat the word five more times and then the process is repeated with the next item. If incorrect, the SLP presents a minimal pair word. If production of the minimal pair is correct, the SLP then shows the participant the letter representing the target sound and again request a repetition of the word. If correct, the participant is asked to repeat it five times and then moves to the next item. If not, additional cues are provided including integral stimulation is used in which the participant is requested to "watch me, listen to me, and say it with me" and then articulatory placement cues. If, after all cues, the participant still cannot produce the target word, he is moved to the next item.

Treatment occurred from 9:00 AM- 12:45 PM, weekdays, for 2 weeks resulting in a total of 30 treatment hours, after accounting for probes and one 15-min break. The first list was presented orally in blocked fashion meaning that all of one sound from a list was presented, then the second, the third, and then back to the first. In order to replicate Wambaugh et al. (2013), as precisely as possible, the second list was presented randomly so that there was mixed presentation of the three of the sounds on that list. The third list was presented blocked; the fourth randomly. The goal was repetition of the target word given cueing according to the SPT Treatment Hierarchy (e.g., Appendix E in Wambaugh et al., 2013). The participant was occasionally provided with positive feedback to reinforce his effort during treatment and also to confirm that he was making progress as he became very competitive with himself and wanted to produce all words in a list.

In order to ensure treatment fidelity, the two graduate student clinicians providing treatment were trained on the procedure by thoroughly reviewing the above-mentioned treatment materials and then practicing it with the PI (first author) prior to client treatment. To ensure that the treatment regimen was followed as trained, the PI also observed 50% of all sessions. In addition, both of the treating graduate student clinicians were in the room with the client at all times. When one was treating, the other was preparing for treatment and observing. The rigidity of this treatment protocol and the presence of other clinicians held those providing treatment to a high standard and helped to ensure that SPT was conducted exactly as described by Wambaugh and colleagues (nd).

Analyses

Two raters (the third and fourth authors) who were seated behind one-way glass so as not to distract from treatment scored the accuracy of each production elicited during probe testing. Since productions were slow and monosyllabic, raters were able to transcribe responses phonetically in real time. Correct articulation of the full word was necessary in order to be scored as a correct production. Any sound distortion meant the word was considered inaccurate. If the participant self-corrected, the corrected word was counted. For example, "kas, no, gas." If self-corrections were inaccurate, raters were to score the response as inaccurate even if the initial word was correct "Gas, no kas." There were no instances of inaccurate self-corrections, however. Scoring was done independently and then compared between raters. Disagreements were resolved via review of the audio recordings with the PI (first author). Effect sizes (ES) were calculated (mean of the baseline probes subtracted from the mean of final two probes divided by the standard deviation of the baseline scores) for trained and untrained items during the treatment phase and the follow-up period and compared to SPT benchmarks (Bailey, Eatchel, & Wambaugh, 2015). Effect sizes were calculated for maintenance by comparing performance at baseline to performance at follow-up.

Audio recordings were used to transcribe the responses from baselines, every third probe, and follow-up probes using broad IPA transcription to analyze errors. All consonants produced within the 144-word stimuli (24 trained and 12 untrained per list) were analyzed and assessed as being produced either accurately or inaccurately. Total accurate productions were divided by the total attempted productions to calculate percent accuracy for each manner of articulation (stop, fricative, affricate, nasal, lateral, or retroflex).

Finally, the two raters also recorded the total number of attempted self-corrections – the number of accurate self-corrections plus inaccurate self-corrections. They then calculated the percentage of all productions that included attempts and the number that included only *correct* attempts at self-correction. All instances of self-correction were patient initiated; these were never encouraged nor discouraged at any time throughout treatment. Any rater disagreement was resolved by reviewing the audio tape. Mark's verbal productions were slow and deliberate and nearly every self-correction was preceded with the word "no" to indicate his awareness that the first production was incorrect. As a result, rater disagreements were rare.

Results

Treatment responsiveness

Single-subject design data for correctly articulated words in each list are shown in Figure 1. Each graph within the figure depicts accuracy for that list over 2 weeks with the treatment

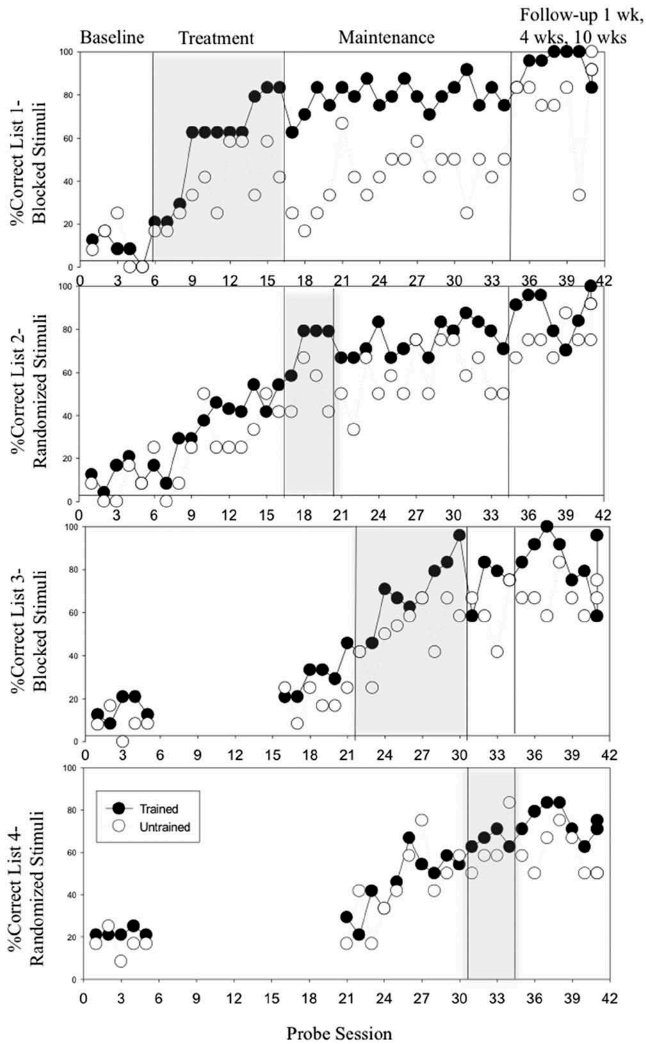

Figure 1. Accuracy of production of probe words.

period shaded in gray. Where treatment ends in one graph, it begins in the next. Criterion was met on three of the four lists with 80% accuracy on three consecutive trials by the end of the two-week period. Of note, increases on lists 2–4 were noted during probes prior to the actual treatment. Maintenance data are shown after the treatment period for each list. Follow-up probes were recorded 1, 4, and 10-weeks post-treatment.

Mark demonstrated improvement in accuracy of production for both treated and untreated words with continued improvement *after* treatment completion for the first two lists. Effect sizes were calculated and compared to benchmarks based on 24 SPT recipients (Bailey et al., 2015). Benchmarks were provided for treatment effect calculated with baseline compared to final probes of the treatment phase (small-5.23, medium-6.98, large-9.65) and for a maintenance effect which was baseline compared to follow-up probes (small-5.9, medium-7.12, large-10.19). For the treatment phase, Mark showed a

Table 2. Effect sizes (ES) for each list.

	Treated Items		Untreated Items	
Stimuli	Treatment Phase ES	Follow-up Phase ES	Treatment Phase ES	Follow-up Phase ES
List 1	11.4	14.1	3.4	6.2
List 2	9.3	10.9	7	10
List 3	10.6	11.6	8.3	9.9
List 4	10.8	12.8	8.3	6.9

Note: Benchmarks according to Baily (2015) are as follows. Treatment phase: *Trained Items* – small– 5.23, medium– 6.98, large– 9.65; *Untrained Items* –small– 2.25, medium– 3.75, large– 6.66. Follow-up phase: *Trained Items* –small– 5.9, medium– 7.12, large– 10.19 for treated materials. *Untrained Items* – small– 2.59, medium– 4.23, large– 6.47.

medium to large ES of 11.4, 9.3, 10.6 and 10.8 for each of the four lists, respectively. Large ESs for the maintenance period were calculated at 14.1, 10.9, 11.6, and 12.8. Gains were maintained and increased in some cases. Performance declined for lists two-four at the 4-week follow-up but recovered at the 10-week post-follow-up.

Benchmarks are also provided for untrained items in the treatment phase (small– 2.25, medium– 3.75, large– 6.66) and follow-up phase (small– 2.59, medium– 4.23, large– 6.47) (Bailey et al., 2015). According to these, medium to large effects were observed across the board for Mark. (treatment phase-3.4, 7.0, 8.3, 8.3; follow-up phase-6.2, 10.0, 9.9, 6.9). See Table 2.

The ABA-2 was administered pre- and post-treatment to quantify potential change in AOS severity but the participant was unable to complete most of the items and a total score could not be calculated. (See Table 3 for scoring on completed subtests.) As a result, this test could not be used as evidence of change. Many of the subtests result in raw scores that increase with the number of errors. For example, in Subtest 2A, words of increasing length are modeled and a repetition is required. If a client accurately repeats "thick" but produces errors with "thickening" this would be scored as a sign of deterioration. A total deterioration score would indicate more severe AOS. Mark, however, could not produce most of the words resulting in a low deterioration score (0–0 = 0) suggesting a less severe AOS if the raw score was to be interpreted without context. This was the case for most of the ABA-2 subtests, therefore total scores were not calculated. Consequently, for this individual, the ABA-2 test was not an accurate measure of change in AOS severity. That said, the participant's improvements in overall speech productivity on the test might be useful as an index of treatment generalization to untrained materials. For example on Subtest 2A, the participant was asked to repeat words of increasing word length (i.e., thick, thicken, thickening). He scored a total of 2 points on the list of shortest words prior to treatment. This increased to 13 immediately post-treatment and to 17 points at follow-up. Increased speech productivity was also noted on Subtests 4, 5 and 10 but these were more difficult and thus the increases less marked.

Aphasia severity

The WAB-R AQ was administered pre and post-treatment to quantify any potential change in aphasia severity. On the WAB-R AQ, repetition, naming, and sentence completion improved from pre to post-treatment but a clinically significant increase (≥ 6; Hula, 2010) was not observed on the WAB-R AQ until the 10-week follow-up (see Table 3). At

Table 3. Pre- and post-treatment WAB AQ, CETI, ABA scores.

Test/subtest (total points)	Pre-treatment	Post-treatment	Follow-up I (4 weeks)	Follow-up II (10 weeks)	Max change pre-treat-ment-Follow-up II
CETI (partner rating)	69.5	n/a	100	101	31.5
CETI (self-rating)	46	n/a	62	78.5	32.5
WAB AQ (100)	27.4	30.9	30.8	39.1	11.7
*ABA**	n/a	n/a	n/a	n/a	n/a
		WAB AQ Subtests			
spontaneous speech (20)	3	3	3	5	2
aud. verbal comprehension (10)	7.5	7.55	7.4	8.15	0.65
repetition (10)	0.9	3	2.3	3	2.1
naming and word finding (10)	2.3	3.1	2.7	3.4	1.1
object naming (60)	20	21	17	24	4
word fluency (20)	0	1	1	2	2
sentence completion (10)	3	6	6	4	1
responsive speech (10)	0	3	3	4	4
		ABA Subtests			
ST1: Diadochokinetic rate	0	0	0	0	n/a
ST2A: Increasing Word Length	2	11	11	9	n/a
ST2B: Increasing Word Length	0	5	Inc**	2	n/a
ST3A: Limb Apraxia	48	42	48	46	n/a
ST3B: Oral Apraxia	42	40	43	35	n/a
ST4: Latency Time and Utterance Time for Polysyllabic Words	100 and 100	100 and 100	100 and 100	74 and 92	n/a
ST5: Repeated Trials	0	0	Inc**	0	n/a
ST6: Inventory of Articulation Characteristics of Apraxia	12	14	Inc**	32	n/a

Note: CETI-Communicative Effectiveness Index; WAB-AQ-R- Western Aphasia Battery Aphasia Quotient Revised; ABA – Apraxia Battery for Adults. * Except on Subtest (ST)1, 3A, and 3B, larger numbers indicate more errors on the ABA. In the case of MARK, who had very little productivity, there were more errors in repeated test conditions but also more overall oral-verbal production. Given MARK's inability to complete much of the testing, an overall ABA score was not calculated. **Inc. refers to subtests that were not completed due to client frustration.

this time, the score was 11.7 points higher than pre-treatment with maintained gains in repetition and increases on auditory comprehension and naming subtests.

Mark scored a 46 on the Communication Effectiveness Index (CETI) at baseline. This increased to 62 at follow-up I and to 78.5 at follow-up II. Questions concerning responsiveness and participation were those that increased the most. Spouse's ratings are often inconsistent with patient and clinician scores but Lomas and colleagues remark that this is irrelevant because the focus of the CETI is on the *change* score (Lomas et al., 1989). In this case, the spouse rated higher than the patient with a 69 pre-treatment, 100 at follow-up I, and 101 at follow-up II. The changes reported by Mark and his spouse were more consistent at 32.5 and 31.5, respectively.

Mark's ratings indicated large positive changes in getting someone's attention; having a one-to-one conversation; and indicating he understands what is being said to him. His spouse's ratings indicated that largest changes were in getting involved in group conversations that are about him; responding to or communication; having a one-to-one conversation; and participating in conversations with strangers. Post-treatment, both Mark and his spouse assigned the lowest ratings to describing something in depth and in having a spontaneous conversation and/or changing the subject with both familiar and unfamiliar listeners.

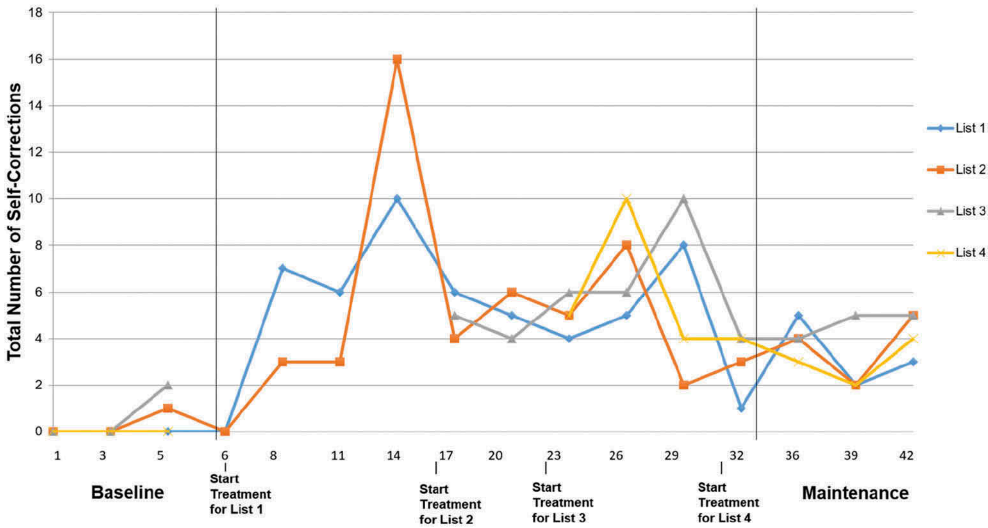

Figure 2. Total number of attempted self-corrections of trained and nontrained words.

Self-corrections

Total correct and incorrect responses were calculated. Of those, the number of attempted self-corrections was noted as a percentage of the total (Figure 2) and as a percentage of incorrect responses only (Figure 3). In addition, successful attempts as a percent of all opportunities were noted and then further broken down by manner of articulation (Figure 4). For lists one-three, it appears that self-correction attempts

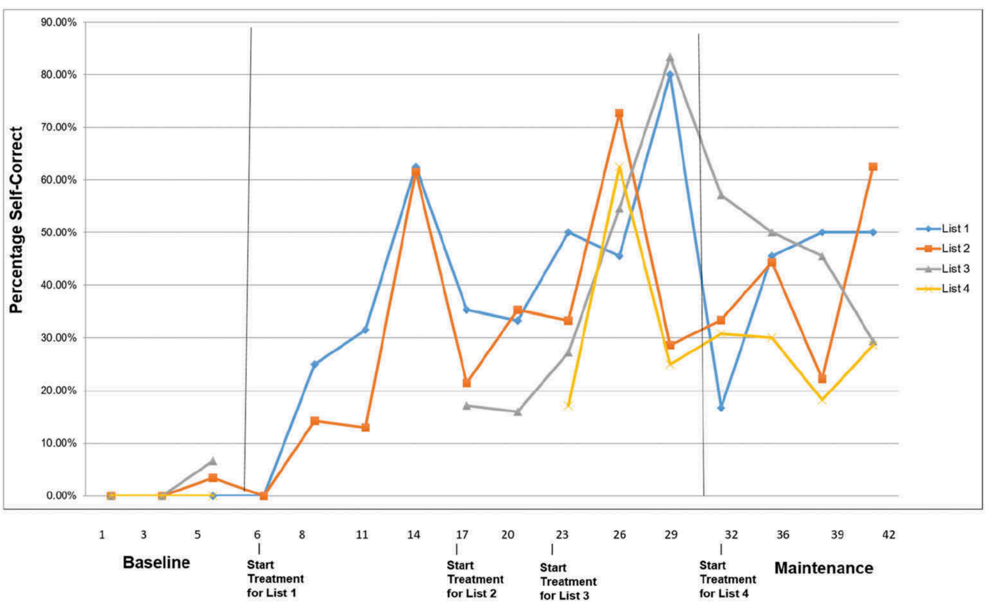

Figure 3. Attempted self-correction out of the total number of incorrect productions.

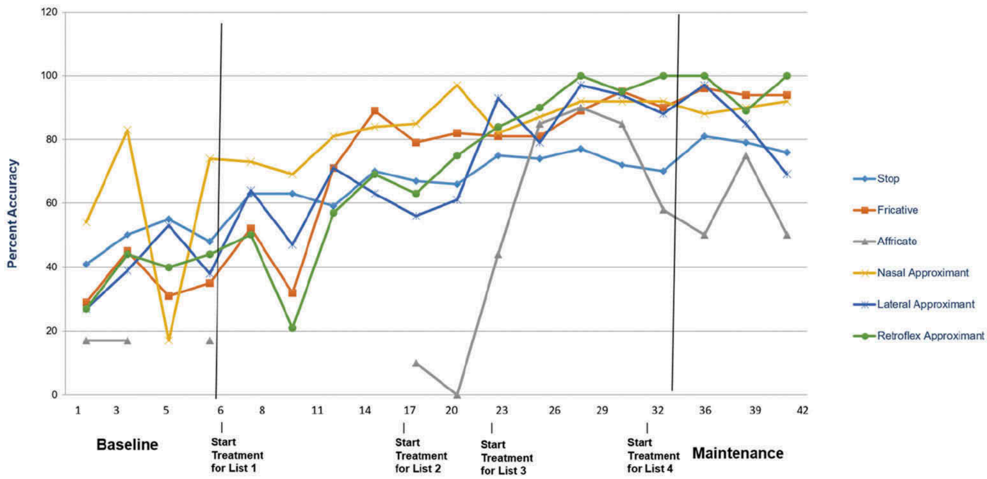

Figure 4. Accuracy by manner of articulation.

increased just to the point of mastery of the material and then decreased in response to overall improvements in accuracy (See Figure 2). The ratio of self-correction attempts to opportunities for self-correction remained the same, however. In list four, self-corrections peaked well before mastery. Pre-treatment, over three baseline probes, a total of three self-correction attempts were made, none of which were accurate. This total number of self-correction attempts increased to a maximum of 29 at probe 26. By that time, 63% of all attempts were successful self-corrections and exceeded the number of unsuccessful self-correction attempts. (See Figure 3.) After the 26th probe, the number of total attempts steadily decreased but this was in proportion to increased accuracy on the first attempt. By follow-up, the number of self-corrections remained stable with an increase in accurate self-corrections and a decrease in inaccurate self-corrections. Overall, the number of correct attempts as a ratio of all attempts increased over time. The percent of attempted self-corrections out of incorrect productions also appeared to increase over time with a dip on the final day of treatment that was not completely recovered at follow-up testing. (See Figure 3).

Errors within each manner of articulation

A total of 3,564 consonant attempts were analyzed. These included 1240 stops, 885 fricatives, 137 affricates, 524 nasals, 477 lateralizations, and 301 retroflex approximations. Increased proficiency in repeating words resulted in additional opportunities to produce each specific manner of articulation. Stop productions were the most practiced manner and yet this did not result in increased gains for that particular manner, as predicted. Rather, *all* manners of articulation improved that were included in probed items. Percent increases from the baseline compared to follow up was calculated for the following: stops- 57.7%, fricatives- 167.1%, affricates- 242.6%, nasals- 58.8%, laterals- 115.9%, retroflex-151.0%. Accuracy became more consistent for all manners as observed in Figure 4.

Discussion

Despite participant heterogeneity across studies, results following SPT tend to be positive and gains generally maintained when the total treatment durations are protracted. In the current study, gains in articulatory accuracy were apparent in a patient with severe AOS after 30 h of treatment conducted over a two-week period. Given the evidence presented in a meta-analysis by Bailey et al. (2015), participants with greater initial severity tend to be associated with higher effect size, particularly for untreated exemplars, therefore we cannot necessarily attribute these effects to the treatment intensity but rather to the low baseline. Additionally, this study is just one person's response to one specific treatment and treatment schedule and patient variability is so great that these findings cannot be interpreted as a method or rigor that would work for all. Despite these caveats, this study does add to what remains a sparse literature.

Mark was someone with very severe AOS who had been working with a speech pathologist three times per week prior to his discharge for lack of gains. He received no other therapy at any time throughout the treatment period (including the follow-up period) and was reported by his wife to be alone for most of the day, while she worked, when he was not participating in the treatment study and for the 10-week follow-up period. It seems likely, then, that the gains realized during the treatment period and afterward can be attributed to the SPT. Gains continued post follow-up and generalized to untreated exemplars. This differed from results reported by Wambaugh et al. (2013) in which gains were not well maintained after 16 h over 4 days or after 16 h over 5.5 weeks (1 h per day, 3 days per week). It was similar, however, to their more recent results (Wambaugh et al., 2018) in which better maintenance was reported. In the current study, gains continued at 10 weeks post-treatment, consistent with reports following this dosage of aphasia therapy.

Intensive treatments are generally thought to confer additional benefits but only if the participant can withstand the rigor. Brady et al. (2016) report higher dropout rates for participants who receive more intensive treatments (2016). In previous studies conducted in our lab, about half of the participants report fatigue after a long block of treatment and may nap afterward. However, all of the participating clinicians observed that Mark was vigilant throughout long, repetitive treatment periods and he reported no additional fatigue afterward. We believe that hearing his own voice for the first time after so long was highly motivating and we that his obvious progress kept him pushing himself throughout the session. In fact, he would often ask to begin again after before the full break-period was complete. His unflagging attention was surprising to those administering the treatment and suggests that intensively administered SPT is a viable option for some individuals.

Aphasia severity, which was monitored but not targeted during treatment, decreased 10 weeks post-treatment. Modest gains in spontaneous language production contributed to this gain. Treatment-induced gains four or more weeks after the completion of treatment have been documented in several aphasia treatment studies using this dosage contributing to a reduction in aphasia severity (Johnson et al., 2013; Maher et al., 2006; Meinzer et al., 2004; Mozeiko et al., 2016, 2018; Pulvermuller et al., 2001; Szaflarski et al., 2008). Since it is not the target of treatment in AOS, aphasia severity is not always assessed post-treatment, but results from the current study raise the question as to whether *any* speech or language stimulation provided at this dosage might serve to decrease aphasia severity by improving ease of communication. It is possible that SPT provided at this alternate dosage may be effective in decreasing aphasia severity. Conversely, it is also possible that providing an intensive *aphasia* treatment may serve to improve AOS severity.

Dose is defined by Warren et al. (2007) as the number of teaching episodes per session which, in this case, translates to the number of times a stimulus is presented and a response elicited. Dose of manner of articulation did not appear to impact proficiency in that manner. Affricates were only presented a quarter of the times stops were presented and yet improvements followed the same trajectory. When manner of articulation was analyzed by Wambaugh et al. (1998), it was determined that sound groups changed in response to treatment but there was little generalization between sound groups. These two studies followed only two individuals but, if they are indicative of responsiveness of others with AOS, it is possible that instead of using lists with specific sound types, that functional lists (of family members names, household items, hobby-related words, etc.) might be developed to serve the same purpose.

The blocked vs. randomized approach was utilized only in order to replicate Wambaugh et al.'s (2013) study as closely as possible. The participant's response to blocked vs. random design is consistent with other reports in that there was a positive response to both. It is not possible to interpret further than this given the short treatment duration.

Mark was unable to produce most words at baseline. As he improved and gained in confidence, he also demonstrated an improved ability to correct speech errors comparable to the results described in the aforementioned study by van der Merwe (2007). The latter study was conducted twice a week for 1 h over a period of 18 months during which there was no increase in the percentage of self-correction per incorrect production. The authors posited that this indicated a continued dysfunctional process of internal predictive control (Van der Merwe, 2007). If so, it is possible then that in the current study in which treatment was provided for 3 h per day, 5 days per week for 2 weeks, and the percentage of self-corrections increased, internal predictive control did, in fact, improve. Perhaps more relevant is the fact that self-corrections seem to precede or coincide with later accurate responses, in which case self-corrections might indicate that gains would continue if treatment was to continue. Self-corrections declined at follow-up, particularly for lists 3 and 4 despite maintained production ability. It is possible that confidence or lack of practice influenced this skill. Of interest is whether *explicitly* training self-corrections ("Do you want to try again?") would increase the rate at which a patient might achieve accurate initial responses. These data suggest that intensive SPT may be useful in improving (1) the ability to predict speech errors and (2) volitional control of speakers with AOS.

Study limitations

Limitations to this study preclude generalization of findings to the larger population of individuals with AOS. Most critically, this pilot work should be applied to a larger participant group. A comparison of the same treatment provided at this same schedule compared to different durations or frequencies would aid interpretation of findings.

Given that a patient might be most responsive to the first treatment he or she receives, the authors would not recommend extending the treatment and manipulating dosage parameters for the same patient. It would be better if there were two groups receiving the same treatment with manipulation of one dosage parameter.

An additional limitation is the appearance of loss of experimental control given the rising probes prior to treatment of lists 2–4. At initial baseline, pre-treatment, the participant could repeat 0–5 single syllables from each list of 24 stimuli. His frustration was palpable during this phase and the investigators thought it likely that treatment would need to be discontinued in order to offer something where he could experience more success. By the fourth hour of treatment, he was consistently repeating 60% (14/24) of the stimuli and was clearly pleased and relieved to experience this success. He repeated newly learned words to himself during breaks and on the car ride home, as reported by his wife. His starting point was so low that the practice of a final consonant in list one (/k/, for example) may have served as practice for the same initial consonant in a later list. In any case, the probes of the yet-to-be-trained lists continued to increase prior to treatment. This is a desirable clinical outcome but may lead researchers to assume a loss of experimental control, perhaps an assumption that the gains are occurring due to some extraneous variables for which we have not accounted. Given the patient history, his lack of activity outside of the treatment study and the fact that Mark had been previously discharged from therapy due to lack of gains, we do believe that he was responding to the intensive SPT administration. It should be noted that the process of probing was a request for repetition, just like the treatment. Therefore, some practice effect is not surprising during the period prior to the exclusive focus on that list. These factors must be considered but should not detract from the fact that Mark increased his repertoire from ~10 repeated words to >100. That said, future studies should include a measure in which gains would not be anticipated to serve as a control probe.

Conclusion

We observed that 30 h over 2 weeks of SPT for an individual with severe apraxia of speech and aphasia resulted in improved accuracy of trained repeated productions, untrained equivalent exemplars, increased self-corrections, and also reduced aphasia severity. We saw no benefit to an increased dose (number of repetitions) of a specific manner of articulation. Importantly, and perhaps most provocatively, maximum gains were observed not immediately post-treatment but rather 10-weeks post-treatment, despite no treatment or other variation from his schedule of staying at home alone while his wife was at work. This finding is consistent with others following this same treatment schedule, lending additional credence to this dosage schedule and warranting investigation of the consolidation process.

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Disclosure statement

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Data Availability

Standardized test scores and all data used to support the findings of this study are included within the article.

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