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IMPROVEMENTS IN SPEECH OF CHILDREN WITH APRAXIA:
THE EFFICACY OF A TREATMENT FOR ESTABLISHING MOTOR PROGRAM
ORGANIZATION (TEMPO)

BY

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B.S. Chemistry, B.A. Spanish; University of New Hampshire, 2013

THESIS

Submitted to the University of New Hampshire
in Partial Fulfillment of
the Requirements for the Degree of

Master of Science
in
Communication Sciences and Disorders

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TABLE OF CONTENTS

	PAGE
ACKNOWLEDGEMENTS.....	iv
LIST OF TABLES.....	v
LIST OF FIGURES.....	vi
ABSTRACT.....	vii
INTRODUCTION.....	1
METHODS.....	6
RESULTS.....	14
DISCUSSION.....	18
REFERENCES.....	27
APPENDIX A: WITHIN-SUBJECT DATA.....	32
APPENDIX B: INSTITUTIONAL REVIEW BOARD APPROVAL.....	33

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LIST OF TABLES

- 1 Principles of motor learning
- 2 Participant and group characteristics
- 3 Post-hoc group*time comparisons for intersegment duration, corrected for multiple comparisons
- 4 Post-hoc group*time comparisons for PVI(dur) in SW stimuli, corrected for multiple comparisons
- 5 Post-hoc group*time comparisons for PVI(dur) in WS stimuli, corrected for multiple comparisons

LIST OF FIGURES

- 1 Definition of time points for treatment and delayed treatment groups
- 2 Mean intersegment duration (a) for combined groups over treatment phase and (b) by group over time Lexical stress measures
- 3 PVI(dur) values for the combined groups across treatment phase for (a) SW stimuli and (b) WS stimuli; group means at Time 0, 1, 2, 3 for PVI(dur) for (c) SW stimuli and (d) WS stimuli.
- 4 Generalization of treatment effects to untreated stimuli

ABSTRACT

IMPROVEMENTS IN SPEECH OF CHILDREN WITH APRAXIA: THE EFFICACY OF A TREATMENT FOR ESTABLISHING MOTOR PROGRAM ORGANIZATION (TEMPO)

by

Hilary E. Miller

University of New Hampshire, May, 2018

Childhood apraxia of speech (CAS) is a motor speech disorder characterized by distorted phonemes, segmentation (increased segment and intersegment durations), and impaired production of lexical stress. This study investigated the efficacy of Treatment for Establishing Motor Program Organization (TEMPO) in nine participants (ages 5 to 8) using a delayed treatment group design. Children received four weeks of intervention for four days each week. Experimental probes were administered at baseline and post-treatment—both immediately and one month after treatment—for treated and untreated stimuli. Significant improvements in specific acoustic measures of segmentation and lexical stress were demonstrated following treatment for both the immediate and delayed treatment groups. Treatment effects for all variables were maintained at one-month post-treatment. These results support the demonstrated efficacy of TEMPO in improving the speech of children with CAS.

Introduction

Childhood apraxia of speech (CAS) is a motor speech disorder characterized by an impairment in the programming of spatial and temporal parameters for accurate speech movement patterns (American Speech-Language-Hearing Association [ASHA], 2007; McNeil, Robin, & Schmidt, 2009). Specifically, apraxia of speech is a breakdown in the translation of intact linguistic and phonological plans into a motor program specifying the exact movement parameters required for segmental and prosodic accuracy of speech production (McNeil et al., 2009). Although the perceptual features that define the disorder have long been the subject of debate, there is emerging consensus that childhood and acquired forms of apraxia are unified by a common set of perceptual features including distortion of speech sounds, segmentation (increased segment and inter-segment duration), and de-stressing of stressed words and syllables (McNeil et al., 2009; Murray, McCabe, Heard & Ballard, 2015).

Acoustic measures of the speech of children with CAS provide additional evidence to support these perceptual criteria. Typical speakers alter three acoustic variables to mark stressed syllables: increased vowel duration, vocal intensity, and fundamental frequency (Kager, 2007). Acoustic measures of these variables in children with CAS demonstrate abnormal production of lexical stress patterns (Munson, Bjorum, & Windsor, 2003; Nijland et al., 2003; Shriberg, Aram, & Kwiatkowski, 1997; Shriberg, Campbell et al., 2003; Velleman, & Shriberg, 1999). Acoustic measures also show significantly longer speech segments (i.e. segmentation) in the speech of children with CAS with reduced variability in the duration of speech segments (Nijland et al., 2003; Shriberg, Green, Campbell, Mcsweeny, & Scheer, 2003). Inappropriate prosody, including lexical stress, has been shown to impact speech intelligibility in a range of neurological disorders (Klopfenstein, 2009; Paul et al., 2005).

Multiple experimental paradigms—including sensorimotor perturbations, visuomotor tracking, “self-select” reaction time studies, and computational modeling—provide evidence that apraxia of speech is an impairment in the activation or modification of spatiotemporal parameters of feed-forward commands for individual speech movements (see Ballard, Granier, & Robin, 2000; Ballard, Tourville, & Robin, 2014; Clark & Robin, 1998; Hageman, Robin, Moon, & Folkins, 1994). Apraxia of speech has been conceptualized within a motor programming model in which there is an internal working memory buffer that holds motor units prior to execution, and a sequencer that places those units in the correct serial order after movement onset. Speakers with apraxia demonstrate impairments in the working memory buffer, not the sequencer (Maas, Robin, Wright & Ballard, 2008). As motor learning occurs, individual speech units are concatenated into larger motor programs; however, this process is impaired in speakers with apraxia as the more complex program places an increased load on the memory buffer (Maas, Robin, Wright, & Ballard, 2008). Specifically, inefficient concatenation explains the perception that individuals with apraxia insert pauses between syllables, or segment their speech. Concatenation also allows for accurate programming of coarticulatory effects and prosodic patterns across syllables, both of which are notably impaired in speakers with apraxia of speech (see Maas, Robin, Wright, & Ballard, 2008). The application of the suprasegmental features that underlie lexical stress (e.g., changes in frequency, intensity and duration) occurs in the working memory buffer so that impairments in this programming stage result in the equal stress patterns observed in apraxia.

Children with CAS often require years of intensive therapy, reportedly up to 81% more therapy than severe phonological disorders to achieve similar functional outcomes (Campbell, 1999; ASHA, 2007). Symptoms can persist into adulthood and result in a substantial disability

affecting intelligibility, social communication, academic performance, and overall quality of life (Carrigg, Baker, Parry, & Ballard, 2015; Rusiewicz, Maize, & Ptakowski, 2017). Although there are a variety of treatment approaches (see Murray, McCabe, & Ballard, 2014 for a review), the efficacy of each has not been established. Treatment approaches for CAS have primarily targeted improved accuracy of segmental features to expand phonemic inventory or develop a core vocabulary, instead of addressing the underlying impairment and the resulting disruption in temporal control of syllable-level prosody —specifically production of lexical stress contrasts— that characterizes the disorder. Consequently, there is a critical need for development and implementation of innovative treatment approaches that effectively target the underlying impairment in CAS.

Motor learning provides a framework both to understand the disorder mechanism and to guide intervention design so as to maximize treatment efficacy. Developed within the framework of Schema Theory of Motor Control and Learning (Schmidt, 1975), Principles of Motor Learning (PML) describe a set of practice and feedback conditions known to promote relatively permanent changes in the capability for movement (Schmidt, & Lee, 2005). Broadly, PML describe important factors in stimulus selection, practice structure, and feedback that increase the difficulty of trained skills and encourage self-evaluation of productions, in order to promote retention and generalization of trained motor skills (Maas, Robin, Austermann Hula et al., 2008; Schmidt & Lee, 2005). In addition, these principles are supported by principles of experience-dependent neural plasticity (Kleim & Jones, 2008; Ludlow et al., 2008). The key principles are summarized in Table 1 (Maas et al., 2008).

Increasingly, treatment literature supports the incorporation of PML in the treatment of CAS (ASHA, 2007; Ballard, Robin, McCabe, & McDonald, 2010; Strand, Stoeckel, & Baas,

Table 1
Principles of motor learning

Condition	Optimal Motor Learning
Practice amount	Higher number of practice trials > less practice
Practice distribution	Distributed practice over longer time period > massed
Practice variability	Variable practice on different targets > constant
Practice schedule	Random practice with intermixed targets > blocked
Attentional focus	External focus on effects of movements > internal
Target complexity	Complex sounds and sequences > less complex
Feedback type	Knowledge of results > knowledge of performance
Feedback frequency	Reduced feedback > constant feedback
Feedback timing	Delayed feedback > immediate feedback

2006; for a review, see Maas, Gildersleeve-Neumann, Jakielski, & Stoeckel, 2014), and in the treatment of acquired apraxia (Austermann Hula, Robin, Maas, Ballard, & Schmidt, 2008; Ballard, Maas, & Robin, 2007; Bislick, Weir, Spencer, Kendall, & Yorkston, 2012; Knock, Ballard, Robin, & Schmidt, 2000). Consistent with motor learning literature, these studies have shown generalization from trained high-complexity targets to less-complex untreated syllables, increased retention with reduced feedback frequency and specificity, and increased retention and transfer with high-intensity, randomized practice (Maas, Robin, Austermann Hula et al., 2008).

The current study investigates the efficacy of Treatment to Establish Motor Program Organization (TEMPO) as an intervention for CAS. Ballard et al. (2010) first demonstrated a significant improvement in acoustic and perceptual measures of lexical stress following treatment using this novel approach. Subsequent studies of this approach have demonstrated positive treatment effects for specific perceptual measures of articulatory and lexical stress

accuracy (Murray, McCabe & Ballard, 2015; McCabe, Macdonald-D’Silva, van Rees, Ballard, & Arciuli, 2014; Thomas, McCabe, & Ballard, 2014).

TEMPO targets the hypothesized underlying impairment in apraxia of speech (i.e. segmentation of speech into individual sound/syllable units) through trained concatenation of nonsense syllables into longer motor units (i.e. three- or four-syllable pseudowords). Additionally, TEMPO explicitly targets each of the three diagnostic features of CAS through repeated practice of accurate speech sounds, fluent transitions between syllables, and syllable stress contrasts in multisyllabic pseudowords. TEMPO is structured within a motor learning framework that fully incorporates all PML (see Table 1) in order to promote retention and transfer of trained skills.

The current study replicates and extends previous work in this area to demonstrate treatment effect for acoustic measures of segmentation (decreased intersegment duration) and lexical stress (increased durational contrast), using a delayed treatment group design in two groups of children. Specifically, the delayed treatment group received only weekly baseline probes during the immediate treatment group’s intervention period.

The primary hypotheses are:

1. Both groups will demonstrate significantly improved performance in acoustic measures of segmentation and lexical stress as a result of treatment, with retention of treatment effects one-month post-treatment.
2. Treatment effects will generalize to untreated exemplars with maintenance up to one-month post-treatment, as indicated by no significant differences between performance on treated and untreated stimuli at any time point.

Methods

Participants

Participating children were referred to our clinic by their current treating speech-language pathologists in response to a recruitment advertisement. The nine participants ranged in age from 5;10 to 8;4, as of the first day of treatment, with more males than females (six and three, respectively). Age and gender of the nine participants, as well as group assignment and language scores from administration of the Clinical Evaluation of Language Fundamentals- Fifth Edition (CELF-5; Wiig, Semel, & Secord, 2013) are presented in Table 2. All nine participants had normal hearing and no orofacial structural abnormalities, muscle weakness, or altered muscle tone or reflexes. Consistent with McNeil et al. (2009) and Ballard et al. (2010), diagnosis of CAS for each child was confirmed by unanimous agreement amongst experienced members of the research team during the speech tasks of the Motor Speech Examination (Duffy, 2005) based on

Table 2
Participant and group characteristics

		Treatment Group					Control Group					
		04	06	07	08	Group Mean	01	03	12	15	16	Group Mean
Age		6;11	5;10	7;8	8;4	7;2	6;7	6;11	7;0	7;5	7;1	7;0
Gender		M	M	M	F		F	F	M	M	M	
CELF-5 Receptive Language	Sentence Comprehension	4	14	2	7	7	10	14	8	8	12	10
	Word Classes	6	9	7	12	9	9	6	8	8	12	9
	Following Directions	6	7	5	12	8	11	7	5	9	9	8
	Index Score	73	100	69	102	86	100	94	80	89	104	93
CELF-5 Expressive Language	Word Structure	5	9	4	6	6	7	10	10	8	7	8
	Formulated Sentences	3	9	6	10	7	13	8	5	7	9	8
	Recalling Sentences	6	6	4	9	6	13	4	9	8	8	8
	Index Score	69	89	70	90	80	106	85	89	87	89	91
Core Language Score		70	96	66	87	80	102	93	87	86	93	92
Language Content Index		72	96	78	110	89	100	84	80	98	100	92

the presence of the three following features of CAS: distortions, segmentation, and equal syllable stress. Children varied in severity across these three features. Children were all native speakers of English and had no other developmental, neurological, genetic or speech disorders. All were enrolled in speech therapy up until the start of participation in this study and had received speech therapy for at least two years prior to enrollment.

Stimuli

Treatment and experimental probe lists consisted of three-syllable pseudowords (e.g., TAGibu or giTAbu; see Ballard et al., 2010), with either a strong-weak (SW) or weak-strong (WS) pattern over the first two syllables. A list of 72 possible CVCVCV combinations containing three different plosive consonants (/b/, /t/, and /g/) and three different long vowels (/a/, /i/, /u/) was generated, in both SW and WS stress conditions. Of those, 20 syllable strings were randomly selected for treatment (Set 1) in both SW and WS stress conditions. The remaining 18 combinations were left untreated (Set 2) to measure transfer to similar but untreated exemplars of both stress conditions.

Additional sets of treated and untreated four-syllable pseudowords (e.g. BIgutaga and biGUtaga) were created for one participant in the treatment group due to high level of performance on three-syllable stimuli in baseline probes and the initial treatment session.

Experimental Design

This study employed a delayed treatment design, in which participants were pseudo-randomly assigned to either an immediate treatment or delayed treatment group, based on family availability for one of the two treatment sessions. The immediate treatment group received

treatment during the first four-week session, while the delayed treatment group was held at baseline for these four weeks, before receiving treatment in a second session. Participants completed two to six baseline tests to measure pre-treatment performance on each of the stimuli sets. Additional experimental probes were completed during the treatment period, immediately post-treatment, and at one-month post-treatment to measure treatment effects, generalization to untreated items, and retention. Figure 1 shows the timing of experimental probes for both groups. The two groups are relatively well matched for age, gender, and language scores (see Table 2).

Baseline and post-treatment probes contained a total of 120 items, including 20 items (10 SW and 10 WS) randomly selected from Set 1 and Set 2, as well as similar fricative pseudowords, real words containing treated sounds, and a set of more and less complex stimuli (four-syllable versus three-syllable targets) in case a child needed to move up or down a complexity level for treatment, as was the case for one participant whose treatment set contained four-syllable stimuli. Only data from Set 1 and Set 2 are reported here. Four variations of the probe lists were used, such that each child received each version of the list an equal number of times. Lists were presented in a different random order each time they were administered. Stimuli were presented in one of ten randomly selected carrier phrases (e.g., “There’s my _____” or “She has a _____.”).

Each child completed two or three baseline probes before the beginning of the first

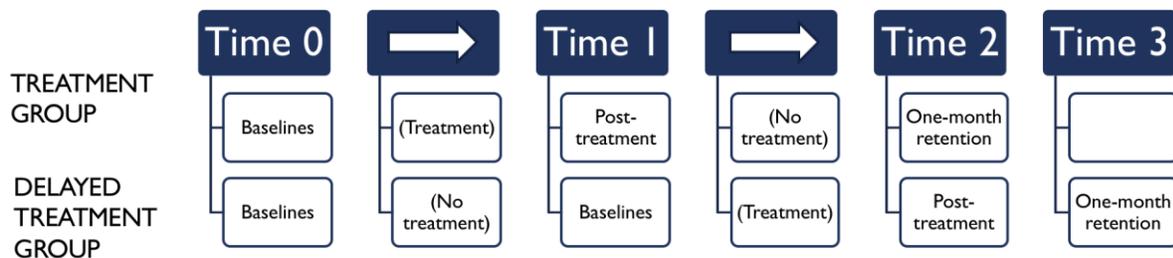


Figure 1: Experimental probe time points for treatment group (top) and delayed treatment group (bottom)

treatment session (Time 0). These were scheduled so as to be completed within about one week, with the last baseline completed about one week before the beginning of the first treatment sessions. Participants in the delayed group completed additional baselines approximately weekly during the first treatment session, for a total of two or three additional baselines by Time 1.

Experimental probes were administered to both groups immediately prior to the first treatment session of the second, third, and fourth weeks. These probes were shortened to 80 items, to allow for completion during the regularly scheduled treatment session. Data from these probes are not reported here.

Two post-treatment probes were completed for participants in both groups. The first, was completed immediately post-treatment ($M = 1.7$ days post-treatment, $SD = 1.6$ days). The second, to measure retention of treatment effects, was completed at approximately one-month post-treatment (average $M = 31.4$ days post-treatment, $SD = 2.2$ days). Children did not receive any speech therapy during this one-month retention phase.

Trained graduate student clinicians administered all baseline probes. Each child's treating clinician administered at least one of the child's baseline probes and all subsequent experimental probes, including both post-treatment probes.

Treatment

Each child was administered treatment by one graduate student clinician. Treatment sessions were conducted four consecutive days a week in 60-min sessions for a four-week period (note that previous work used only a three-week period). At least 50% of each child's sessions were directly observed by the author, while a second experienced investigator also observed a minimum of 25% of each clinician's sessions to ensure inter-rater reliability. Any discrepancies

with treatment protocol were addressed during or between sessions to maintain treatment fidelity. All sessions were also audio and video-recorded.

Intervention explicitly targeted each of the three features of CAS through repeated productions of multisyllabic pseudowords (e.g. tabigu) at a natural speech rate, where correct production was assessed on accuracy across each of the three features of CAS: correct sounds, fluent transitions between syllables, and accurate lexical stress. Twenty stimuli (10 SW and 10 WS) from Set 1 were randomly selected for each session. Treatment was structured within a motor learning framework (Schmidt & Lee, 2005; Maas, Robin, Austermann Hula et al., 2008). Each treatment session consisted of Pre-Practice, continuing until the child correctly produced five stimuli with clinician-provided Knowledge of Performance (KP) feedback and cues as necessary; and Practice, consisting of 100 total productions of the twenty randomly ordered stimuli (see Ballard et al., 2010). The Practice stage adhered to a strict low frequency, delayed feedback schedule with Knowledge of Results (KR) feedback provided on 60% of trials after a 3-second delay. Clinicians used a feedback sheet containing a visual of the three targeted features (sounds, stress, segmentation) during Pre-Practice and Practice to refer to each term as they gave feedback (e.g. “Nice and smooth, but sounds and stress weren’t right.”) There was also a 5-second delay following feedback before presentation of the next stimulus.

For the first two sessions, all children completed an hour of Pre-practice. In subsequent sessions, Pre-practice lasted no more than 15 minutes. Since not all children could read fluently, stimuli were presented auditorily by the clinician, with a 3 second delay between the model and the child’s production.

Equipment

All experimental probes and treatment sessions were recorded in a quiet room at 44.1 kHz with Samson XPD1 microphones, positioned 5 cm from child's mouth.

Acoustic Measures

Acoustic analyses were completed by acoustically-trained research assistants using Praat signal-processing software (Boersma & Weenink, 2001). Lexical stress was measured through comparison of vowel duration (ms) of strong and weak syllables in treated and untreated plosive stimuli on pre- and post-treatment experimental probes. Vowel duration was measured between the first and last glottal pulse of the vocalic nucleus, as indicated by energy extending through F1 and F2 displayed on the wideband spectrogram, and using fundamental frequency, formant, and intensity contours generated by the Praat software (Ballard et al., 2010; Kent & Read, 1992).

The pairwise variability index (PVI) of each variable was calculated using Equation 1 to provide a normalized comparison of the strong and weak syllable in each stimulus:

$$PVI = 100 \times [d_k - d_{k+1}] / [(d_k + d_{k+1}) / 2], \quad (1)$$

where d is the duration of the k^{th} syllable (Ballard et al., 2010; Low, Grabe, & Nolan, 2000). A higher PVI value reflects increased contrast in lexical stress, whereas a PVI of zero indicates equal stress across syllables.

PVI was only calculated for stimuli in which no syllables were omitted, and both strong and weak syllables had a measurable vowel (i.e. not whispered). Stimuli in which the child did not repeat the intended stress target (i.e. produced a WS pseudoword instead of SW) were also excluded from analysis.

Segmentation was measured as intersegment duration, or the amount of time between syllables. This was defined as the time from the last glottal pulse, as indicated by the end of F1 and F2 in the wideband spectrogram, to the onset of the plosive burst in the following syllable.

For items in which children added an extra syllable, only the syllables that best fit the intended stress pattern were included for analysis. For some items, there was no way to mark an intersegment duration due to either omission or severe distortion of the plosive consonant that made it impossible to distinguish start and end points of segments.

Reliability

Approximately 15% of each rater's samples were rescored by a second rater to calculate inter-rater reliability. Each rater also remeasured a random selection of approximately 15% of their samples. Intra-class correlation revealed a high-degree of reliability for inter-rater comparison of both intersegment duration and vowel duration measures (ISD: $r = .953$, 95% confidence interval .945-.960, $F(551,552) = 21.351$, $p < .001$; Vowel duration: $r = .926$, 95% confidence interval .912-.938, $F(530,531) = 13.494$, $p < .001$), as well as for intra-rater comparisons (ISD: $r = .985$, 95% confidence interval .982-.988, $F(298,299) = 68.294$, $p < .001$; Vowel duration: $r = .927$, 95% confidence interval .902-.946, $F(176,177) = 13.779$, $p < .001$).

Data Analysis

The total number of useable stimuli for each participant varied based on a number of factors including technical issues (e.g. recording failed or background noise), subject cooperation (e.g. yelling or laughing during production), errors in clinician models (e.g. transposing syllables), and missing data points within stimuli (e.g. syllable omissions; see Acoustic Measures

for more detail on inclusion criteria for analysis). Outliers, defined as any values over two standard deviations from the pre- or post-treatment mean for that participant, were also excluded from analysis. On average, we lost approximately 30% of data points, unevenly distributed across participants. Missing data from each participant ranged approximately from 10% to 60%, depending in large part upon the frequency with which children omitted unstressed syllables. This variation in the number of data points for each participant necessitated a mixed model with participant as a random effect to control for missing data, with all other variables fixed (Bell, Ene, Smiley, & Schoeneberger, 2013). A significance level of 0.05 was set a priori for all statistical analyses. Post-hoc testing used the Tukey-Kramer test which adjusted for multiple comparisons.

Baseline stability was analyzed for all nine participants for intersegment duration, PVI(dur) of SW stimuli, and PVI(dur) of WS stimuli using a mixed model ANOVA with one independent variable of time (baselines 1 through 6, as applicable). There was no significant effect for time for either PVI(dur) SW ($F(5, 380) = 1.02, p = .41$) or PVI(dur) WS variables ($F(5, 315) = 0.83, p = .53$). Time was significant for intersegment duration ($F(5, 1819) = 2.79, p = .02$), so the two groups were analyzed separately for this variable only. Time was not significant for the immediate treatment group, $F(2, 523) = 2.89, p = .06$. Time was still significant for the delayed treatment group ($F(5, 1292) = 2.39, p = .04$), but this was accounted for by one subject, who was then removed from the analysis. The result was no significant effect of time ($F(5, 985) = 1.97, p = .08$). These data allowed for the combination of baselines for each group into an average for that timepoint for the subsequent analyses.

Overall treatment effects for the combined nine participants were analyzed using a mixed model ANOVA with random intercept for subject, and fixed factor of phase (average baseline,

post-treatment, and retention). The time analysis examined the immediate versus the delayed control group, using a mixed model ANOVA with a random intercept for subject, and fixed factors of time (see Figure 1 for the four time points) and group (immediate treatment, delayed treatment). It was anticipated that group differences would only be present at time 1, following the first treatment session.

A main hypothesis was that there would be no difference between treated and untreated stimuli sets. The main analysis included only time and group as fixed factors, because we did not have enough power to include set which would have necessitated a three-way interaction. Therefore, only the main effect of set was included in the analysis. The results were $F(1, 3102) = 0.00, p = .99$ for intersegment duration, $F(1, 690) = 0.04, p = .84$ for PVI(dur) SW, and $F(1, 590) = 5.07, p = .02$ for PVI(dur) WS. As there was a significant effect of set for PVI(dur) WS, we examined stimulus set within each group for this variable and found no main effect of set (Group 1 (Immediate treatment): $F(1, 217) = 3.31, p = .07$; Group 2 (Delayed treatment): $F(1, 372) = 2.28, p = .13$); therefore we were able to combine sets for the remaining statistical analysis.

Results

Intersegment Duration

Results of phase analysis for the combined nine participants revealed a significant phase effect, $F(2, 3104) = 99.55, p < .001$. Adjusted means for each treatment phase are plotted in Figure 2a. A positive treatment effect is demonstrated by a decrease in intersegment duration, representing reduced segmentation of speech. Tukey post-hoc test revealed significant differences between baseline and post-treatment ($p < .001$), and baseline and retention treatment

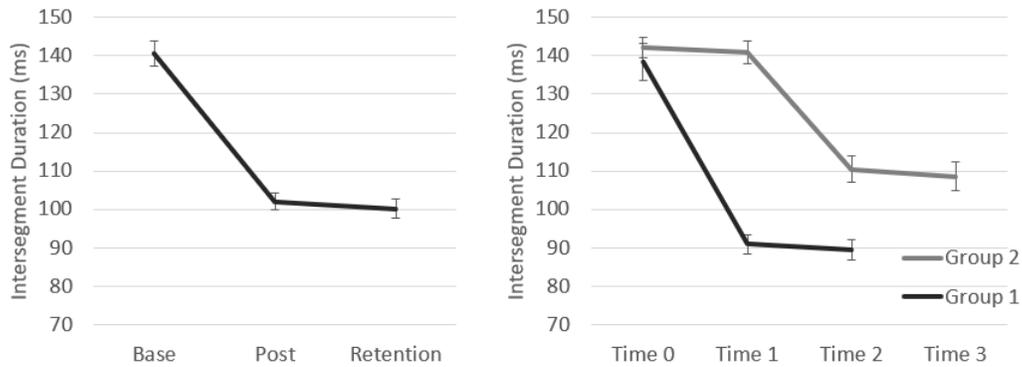


Figure 2: Mean intersegment duration (a) for combined groups over treatment phase and (b) by group over time. All within-group pre- to post-treatment comparisons were significant (see Table 3 for statistical results for all comparisons).

phases ($p < .001$). Immediate post-treatment and retention phases were not significantly different ($p = .94$).

Results for group over time analysis of intersegment duration demonstrate a significant main effect of time ($F(3, 3104) = 64.97, p < .001$) and the interaction between group and time ($F(2, 3104) = 16.52, p < .001$), but the main effect of group was not significant ($F(1, 9.04) = 1.46, p = .26$). The group over time interaction is plotted in Figure 2b. Significant differences were explored using Tukey post-hoc tests (see Table 3 for full post-hoc testing). All baseline time points for both groups were significantly higher than both post-treatment time points ($p < .001$), which did not differ significantly from one another ($p = 1.00$). Group differences were not significant at Time 1 ($p = .35$).

Table 3

Post-hoc group*time comparisons for intersegment duration; adjusted p - values are reported below.

	Group 1, Time 1	Group 1, Time 2	Group 2, Time 0	Group 2, Time 1	Group 2, Time 2	Group 2, Time 3
Group 1, Time 0	<.001	<.001	1.00	1.00	.93	.90
Group 1, Time 1		1.00	.14	.35	.96	.97
Group 1, Time 2			.14	.35	.95	.97
Group 2, Time 0				.15	<.001	<.001
Group 2, Time 1					<.001	<.001
Group 2, Time 2						1.00

Lexical stress

PVI(dur) of SW and WS stimuli were analyzed separately. Results of a mixed model ANOVA revealed a significant phase effect for combined nine participants for both SW and WS stimuli (SW: $F(2, 691) = 29.77, p < .001$; WS: $F(2, 591) = 10.7, p < .001$). Treatment effects are shown in Figures 3a and 3c. Post-hoc tests confirmed significant differences between baseline and post-treatment (SW: $p < .001$, WS: $p < .001$), baseline and retention (SW: $p < .001$, WS: $p < .001$), but no difference between post-treatment and retention (SW: $p = .83$, WS: $p = 1.00$). Mixed model ANOVA of PVI(dur) in SW stimuli revealed a significant main effect for time ($F(3, 691) = 17.62, p < .001$) and the group by time interaction ($F(2, 691) = 7.28, p < .001$). Mean PVI(dur) for each group over time is plotted in Figure 3b. Post-hoc testing showed

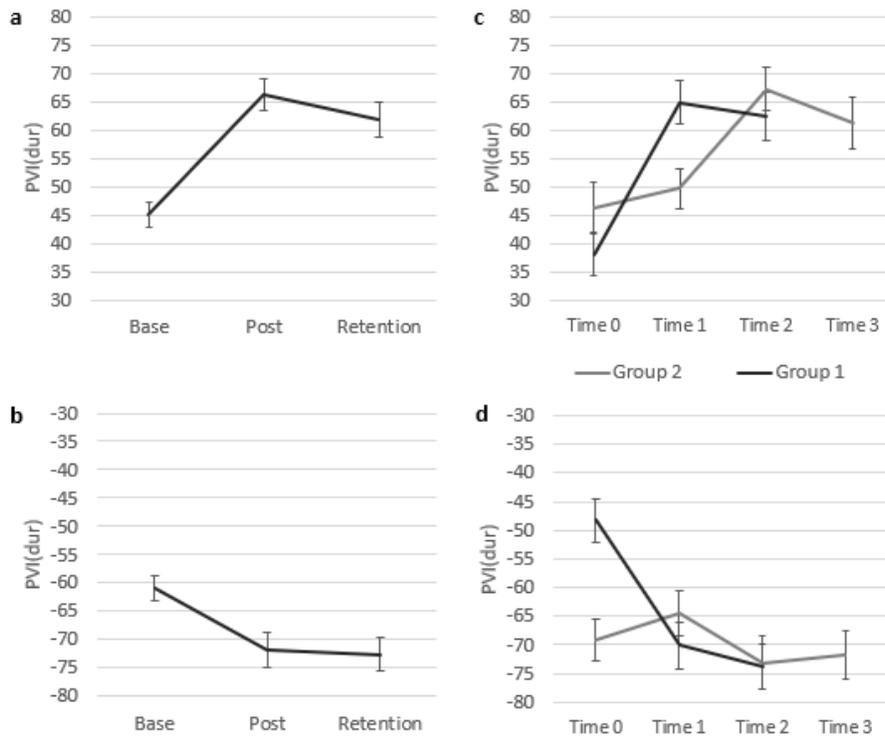


Figure 3: PVI(dur) values for the combined groups across treatment phase for (a) SW stimuli and (b) WS stimuli; group means at Time 0, 1, 2, 3 for PVI(dur) for (c) SW stimuli and (d) WS stimuli. Within-group comparisons between pre- and post-treatment time points were significant ($p < .05$), except for WS stimuli for Group 2 (see Tables 4 and 5 for p -values for all group and time comparisons).

Table 4

Post-hoc group*time comparisons for PVI(dur) in SW stimuli; adjusted *p*- values are reported below.

	Group 1, Time 1	Group 1, Time 2	Group 2, Time 0	Group 2, Time 1	Group 2, Time 2	Group 2, Time 3
Group 1, Time 0	<.001	<.001	.98	.93	.30	.48
Group 1, Time 1		1.00	.88	.96	1.00	1.00
Group 1, Time 2			.89	.97	1.00	1.00
Group 2, Time 0				.97	.001	.04
Group 2, Time 1					.01	.16
Group 2, Time 2						.99

significant differences for both groups before and after treatment (see Table 4 for complete post-hoc testing). Again, between group differences at Time 1 were not significant ($p = .96$).

Group by time analysis of PVI(dur) of WS stimuli also revealed significant main effect for time ($F(3, 591) = 8.86, p < .001$) and group by time interaction ($F(2,591) = 4.31, p = .01$).

There was no difference between the two groups at Time 1 ($p = 1.00$). Post-hoc testing showed significant differences only for Group 1 between the following time points: baseline and post-treatment ($p < .001$), and baseline and retention ($p < .001$). Complete post-hoc testing is summarized in Table 5.

Generalization of Treatment Effects

There were no significant differences between treated and untreated exemplars within

Table 5

Post-hoc group*time comparisons for PVI(dur) in WS stimuli; adjusted *p*- values are reported below.

	Group 1, Time 1	Group 1, Time 2	Group 2, Time 0	Group 2, Time 1	Group 2, Time 2	Group 2, Time 3
Group 1, Time 0	<.001	<.001	.54	.29	.15	.22
Group 1, Time 1		1.00	1.00	1.00	1.00	1.00
Group 1, Time 2			1.00	1.00	1.00	1.00
Group 2, Time 0				.96	.62	.84
Group 2, Time 1					.98	1.00
Group 2, Time 2						1.00

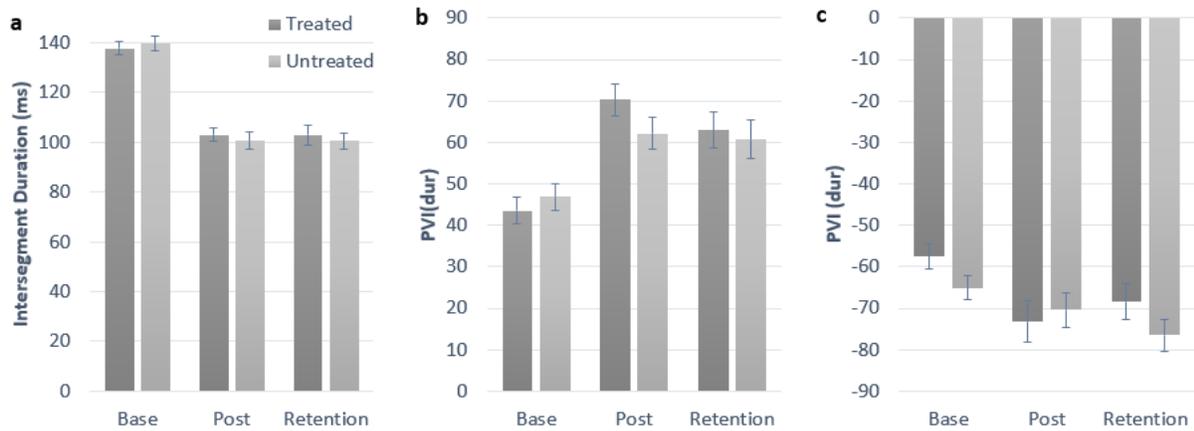


Figure 4: Generalization of treatment effects to untreated stimuli. Plotted are mean acoustic measures (a) intersegment duration, (b) PVI (dur) for SW stimuli, and (c) PVI(dur) for WS stimuli; separated by stimuli set (treated; untreated) for combined groups at each of three treatment phases (baseline; immediate post-treatment; one-month retention). Error bars show standard error of the mean.

either group across the three phases of the study, for any of the three variables (see Methods).

Means for each stimuli set across treatment phases are shown in Figure 4 for the three analyzed variables.

Discussion

This study investigated the efficacy of TEMPO in improving acoustic measures of segmentation and stress using a delayed treatment design with two groups. Critically, both groups demonstrated stable performance across baselines, which allowed for examination of treatment effects. Ideally, we anticipated a significant difference in groups at Time 1 only (following the first treatment period). Despite observed differences in the right direction at that time point, the improvements were not significant at this time point for any of the three variables, likely due to insufficient power as there was high within- and between-subject variability. While the expected group difference at Time 1 was not observed, within-group treatment effects were present for all variables except PVI(dur) in WS stimuli for the delayed treatment group.

Hypothesis 1: Positive Treatment Effects

Results supported the hypothesis that treatment would result in significant improvements in segmentation (intersegment interval duration) and lexical stress for SW and WS stimuli as indexed by PVI from vowel duration measures. In addition, data supported our hypothesis that there would be maintenance of treatment effects at one-month post-treatment. These data replicate previously reported treatment effects and add to the growing literature on the positive effects of TEMPO in remediation of two of the primary features of CAS (Ballard et al., 2010; McCabe et al., 2014; Murray, McCabe, & Ballard, 2015; Thomas et al., 2014). Although McCabe et al. (2014) reported perceptual measures for a relatively large group of participants, this group study is the first to use acoustic outcome measures. Acoustic measures can capture smaller increments of improvement that may not be perceptually measurable. Acoustic measures are a more sensitive measure of change, as they can precede changes in perceptual measures. In addition, acoustic measures provide more fine-tuned detail that allows for provision of an underlying explanation of perceptual measures. A partner study completed in conjunction with this study also demonstrated clinically significant improvements for our nine participants in areas including intelligibility, social communication and play, independence, and overall communicative participation following treatment (Schultz, 2018). In combination, these improvements in specific acoustic variables and global measures of communication provide strong evidence to support the efficacy of TEMPO as an intervention for CAS.

This study also adds to a growing body of evidence supporting the application of PML to the treatment of CAS (Ballard et al., 2010; Maas et al., 2014; Murray et al., 2014). These principles target the structure of sessions including a pre-practice phase before intensive, high-frequency practice. Details of principles critical to success are fully delineated in the introduction

and include practice variables (e.g., random stimulus presentation, high number of trials) and feedback variations (e.g., delayed, reduced frequency, knowledge of results feedback).

Improvements in segmentation

Both the immediate and delayed treatment groups showed reduced segmentation following treatment, demonstrating that TEMPO improves speech motor learning in these children. Children with CAS have a deficit in the working memory buffer that holds motor units (see Maas, Robin, Wright, & Ballard, 2008). Longer or more complex units place too great a demand on the memory buffer; consequently, speech production for these children is limited as they can only hold shorter and less complex motor programs in this impaired buffer prior to execution, resulting in segmentation of speech. As speech motor learning occurs during treatment, shorter segments—such as gi, ta and bu—are combined into multi-syllable units such as gitabu. In essence, smooth speech emerges over the course of intervention because the separate syllable motor programs concatenate into a single larger program. Significant decreases in intersegment duration confirm improvements in speech motor learning.

Improvements in stress contrasts

Interpretation of lexical stress improvements may be limited by the inclusion of only PVI(dur) as a measure of stress, while intensity and pitch also impact perception of stress. Nonetheless, durational contrasts play a dominant role in stress production and perception in typical speakers and may provide the most sensitive measure of lexical stress deficits in children with CAS. Toddlers typically use only durational contrasts to indicate lexical stress, before development of pitch and intensity contrasts (Pollock, Brammer, & Hageman, 1993). Duration is

also the primary variable used by typical children to mark stress, particularly if only one of the three variables is used (Davis et al., 2000). Adult typical speakers also use more dramatic contrasts in duration than either pitch or intensity, for both SW and WS stress patterns (Ballard, Djaja, Arciuli, James, & van Doorn, 2012). In addition, PVI(dur) measures of children with CAS have been shown to correlate well with perceptual ratings of accuracy in stress production (Ballard et al., 2010), indicating that limited durational contrasts may be responsible for the perception of equal stress in children with CAS to a greater degree than other prosodic features. This is logical because CAS is a disruption in temporal control of speech, marked by increased duration and reduced variability in duration of speech segments. Therefore, PVI(dur) may be the best indicator of treatment effect, as it reflects the specific impairment in stress production for children with CAS.

Overall, improvements in production of stress contrasts were less robust than treatment effects for segmentation. For SW stimuli in particular, this finding may be explained by the ceiling effect imposed by the comparatively low PVI(dur) values documented for typical speakers. The combined post-treatment average of 62 is well within the expected typical performance range for age-matched children or adults (Ballard et al., 2012).

Participants demonstrated less dramatic improvements in PVI(dur) measures for WS stress contrasts than SW. The WS pattern may require greater amounts of treatment because it develops later than the SW pattern for typical English-speaking children. Production of SW patterns is mastered around age three, while production of WS stress contrasts continues to develop through at least age eleven (Arciuli & Ballard, 2017; James, Ferguson, & Butcher, 2012). Difficulty with WS words may reflect a trochaic bias resulting from increased exposure to the SW stress pattern in English or the increased physiological demands required for production

of WS stimuli (see Ballard et al., 2012 for a review). The WS pattern may be particularly difficult for children with CAS because it requires more dramatic durational contrasts, with reported PVI(dur) of -120 for typical adults (Ballard et al., 2012). In fact, PVI(dur) values in WS words have been proposed as a diagnostic criterion for acquired apraxia of speech, as these stimuli are more sensitive to subtle impairments in control of temporal contrasts (Ballard et al., 2016).

Participants in this study demonstrated considerable difficulty producing all syllables in WS stimuli at baseline, with frequent omission of the initial weak syllable. This mimics the progression, albeit delayed, of typical speakers. Toddlers frequently omit unstressed syllables (Salidis & Johnson, 1997). At about age three, there is a simultaneous decrease in syllable deletion and increase in the perception of equal stress across syllables (Young, 1991). Further analysis of the frequency of syllable omission might show preliminary progress in production of WS stimuli in children with CAS since inclusion of the weak syllable is a prerequisite to production of appropriate durational contrasts. Children with CAS likely require more than four weeks of intervention to reach typical performance for the increased durational contrasts necessary for WS stimuli.

This study was unique in the inclusion of children with below average receptive language scores. Two participants (04 and 07) had scores more than one standard deviation below the age-normed average for the Receptive Language Index on the CELF-5 (standard score 73 and 69, respectively). These two participants demonstrated substantial improvements in measures of segmentation and durational contrasts in both SW and WS stimuli. Additionally, both children demonstrated generalization and maintenance of treatment effects. This finding is especially important because many children with CAS have concomitant language processing difficulties.

Evidence that children with language impairments can successfully engage in an intensive speech motor programming treatment suggests a broad applicability of TEMPO to a large number of children with concomitant CAS. It may be that the use of nonsense syllables minimizes the demands placed on the language system.

Hypothesis 2: Generalization

As hypothesized, treatment effects were consistent across the two stimuli sets for all three variables, demonstrating generalization to untreated syllable strings with retention one-month post-treatment for both groups.

Pseudowords were selected as treatment stimuli, despite limited functionality, because they allow for increased variability of practice. The removal of linguistic constraints maximizes the number of novel combinations in which to practice the targeted sounds and stress patterns, in accordance with PML. Also, removal of the linguistic system from treatment allows for isolated and targeted focus on the motor programming system—the primary impairment in CAS. The use of nonword stimuli also replicates novel word learning. Critically, practice of nonsense syllable strings trains the reorganization of multiple speech units into a single motor program, as it is this concatenation process that is hypothesized to be impaired in apraxia of speech (Maas, Robin, Wright, & Ballard, 2008). The goal of intervention for CAS is not to practice a specific set of movement patterns, but rather to improve the overall efficiency of motor program organization for more accurate programming of coarticulation and prosodic patterns across units in multisyllabic utterances. Therefore, generalization of treatment effects to untreated stimuli not only indicates treatment efficacy, but also informs the underlying theory and mechanism of action upon which this treatment approach was developed.

Clinical observations suggest similar strong generalization of treatment effects to real words and conversational speech for the three primary perceptual features of CAS. Schultz (2018) also reported clinically significant improvements in parental reports of intelligibility, social communication, and overall communicative participation for the children in this study. These communication outcome data serve as an indicator of transfer of treatment effects to other stimuli and natural environments. In summary, the strong generalization of treatment effects indicates improved motor programming efficiency and supports TEMPO's hypothesized mechanism of action in targeting the proposed impairment in concatenation. Critically, the observed generalization provides strong support for the hypothesis that treatments that successfully target the underlying mechanism improve overall speech production, beyond the specific symptoms or stimuli targeted in treatment sessions.

Limitations and Future Directions

The current study is limited by the high degree of between-subject variation at baseline. Treatment effects for each of the three variables were limited by this variability, which included near-typical performance by some children on each of the three variables. Although TEMPO is designed to target all three features of CAS, the nine participants presented with varied profiles marked by a range of strengths and weaknesses across these features. As a result, improvement varied on each feature, especially for participants who presented with only a mild impairment for a given feature (see Appendix for more detail on individual performance). For example, the majority of participants used appropriate durational contrasts for SW stimuli at baseline, with PVI(dur) within the range of 40-50 reported for typical 7-year-olds (Ballard et al., 2012). Two participants, both in the delayed treatment group, also presented with near typical stress

production for WS stimuli, (about -95 for typical seven-year-olds; Ballard et al., 2012). Two participants presented with perceptually minimal segmentation at baseline, with no significant changes in intersegment duration following treatment. However, there are currently no measures of typical intersegment duration for comparison. Segmentation is a key diagnostic feature of CAS, so an analysis of typical intersegment duration measures in age-matched children should be completed to establish a comparison point and better measure treatment effect in future studies. Analysis of only those participants with more severe impairments in each variable may provide a more representative representation of treatment effects.

Acoustic analysis for this study included only temporal measures of supra-segmental aspects of speech. Future work should include other acoustic measures of stress, including fundamental frequency and intensity contrasts, and measures of segmental accuracy, such as voice onset time or other measures of speech-sound distortions. In addition, future work should include perceptual measures of accuracy across the three targeted features. A more formal analysis of generalization to real words and conversational speech, including intelligibility measures, should also be completed.

This study was also limited by the small number of participants. Nevertheless, combined with the previous studies of TEMPO, there is now a demonstrated treatment effect for variations of this approach in more than thirty participants. This body of work necessitates a future clinical trial with a much larger number of participants. Additionally, future work should investigate the efficacy of TEMPO for more diverse participant groups, include exploration of factors that influence individual response to treatment, and establish ideal dosage and treatment intensity (see Baker, 2012; Manes & Robin, 2012). Continued treatment research should establish recommendations for future clinical applications of this treatment approach. Previous studies of

TEMPO have shown improved learning (i.e. generalization and retention) when TEMPO is administered in a high-frequency schedule with four sessions a week (Thomas et al., 2014). This study extended the treatment period from the twelve sessions (three-week period) used in previous studies, to a 16-session treatment period. Future work should further investigate the effect of these variations in treatment schedule on treatment outcomes. In addition, future studies should investigate the efficacy of subsequent doses of TEMPO, particularly for children with more severe impairments in any of the three features.

A critical next step in CAS research is the use of neuroimaging to establish the efficacy of treatments in normalizing brain networks in children with CAS. Neuroimaging studies of apraxia have primarily focused on adults with the acquired form of the disorder. There remains a critical need to establish differences in brain networks in children with CAS and determine the specific neural systems underlying the disorder. Neuroimaging work should also characterize how the treatment induces neural plasticity ultimately allowing for its optimization.

Conclusions

Acoustic measurements demonstrate overall reduced segmentation and improved stress contrasts for nine participants with CAS following four weeks of TEMPO intervention. All three variables demonstrated significant improvements, generalization to untreated syllable strings, and retention of treatment effects. These results replicate and extend previous studies of TEMPO and support the efficacy of this approach in treatment of CAS.

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APPENDIX A: WITHIN-SUBJECT DATA

Subject	Intersegment Duration			PVI(Dur) SW			PVI(Dur) WS		
	Base	Post	Retention	Base	Post	Retention	Base	Post	Retention
01	141	92	90	34	110	116	-69	-115	-111
03	198	184	189	2	28	22	-47	-69	-51
04	169	132	139	5	33	43	-19	-56	-52
06	86	76	74	50	69	61	-30	-61	-59
07	198	76	70	60	83	91	-52	-70	-84
08	77	74	72	23	71	63	-72	-86	-88
12	127	107	101	72	61	64	-50	-48	-51
15	118	92	80	65	70	69	-94	-83	-75
16	130	92	90	87	68	54	-97	-74	-70

APPENDIX B: INSTITUTIONAL REVIEW BOARD APPROVAL

University of New Hampshire

Research Integrity Services, Service Building
51 College Road, Durham, NH 03824-3585
Fax: 603-862-3564

14-Apr-2017

Robin, Donald A
Communication Sciences & Disorders
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Durham, NH 03824

IRB #: 6627

Study: TEMPO Treatment Study

Approval Date: 04-Apr-2017

The Institutional Review Board for the Protection of Human Subjects in Research (IRB) has reviewed and approved the protocol for your study.

Approval is granted to conduct your study as described in your protocol for one year from the approval date above. At the end of the approval period you will be asked to submit a report with regard to the involvement of human subjects in this study. If your study is still active, you may request an extension of IRB approval.

Researchers who conduct studies involving human subjects have responsibilities as outlined in the document, *Responsibilities of Directors of Research Studies Involving Human Subjects*. This document is available at <http://unh.edu/research/irb-application-resources>. Please read this document carefully before commencing your work involving human subjects.

If you have questions or concerns about your study or this approval, please feel free to contact me at 603-862-2003 or Julie.simpson@unh.edu. Please refer to the IRB # above in all correspondence related to this study. The IRB wishes you success with your research.

For the IRB,



Julie F. Simpson
Director

cc: File