

Research Report

Telehealth delivery of Rapid Syllable Transitions (ReST) treatment for childhood apraxia of speech

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Abstract

Background: Rapid Syllable Transitions (ReST) treatment uses pseudo-word targets with varying lexical stress to target simultaneously articulation, prosodic accuracy and coarticulatory transitions in childhood apraxia of speech (CAS). The treatment is efficacious for the acquisition of imitated pseudo-words, and generalization of skill to untreated pseudo-words and real words. Despite the growing popularity of telehealth as a method of service delivery, there is no research into the efficacy of telehealth treatments for CAS. Telehealth service delivery is associated with compromised audio and visual signal transmission that may affect the efficacy of treatment.

Aims: To conduct a phase 1 efficacy study of telehealth delivery of ReST treatment for CAS, and to discuss the efficacy with reference to face-to-face ReST treatment.

Methods & Procedures: Using a multiple baseline across participants design, five children aged 5–11 years with CAS received ReST treatment four times a week for 3 weeks via video conferencing with Adobe Connect. The children's ability to imitate new pseudo-words, generalize the skills to untreated pseudo-words and real word items, and maintain the skills following treatment were assessed. Both visual and statistical analyses were utilized.

Outcomes & Results: All five children significantly improved with their production of the imitated treated pseudo-word items and significantly generalized to similar untreated pseudo-words and real words. Additionally, two of the children showed significant generalization to imitated phrases with the treatment items. Four of the children maintained their treatment gains up to 4 months post-treatment. Telehealth delivery produced similar acquisition of pseudo-words and generalization to untreated behaviours as face-to-face delivery; however, in the 4 months following treatment, the children showed stable rather than improving speech skills. The intra- and inter-judge reliability was similar in telehealth delivery for face-to-face delivery. Caregivers and clinicians were satisfied with the telehealth treatment.

Conclusions & Implications: This phase 1 study provides promising indications of the efficacy of ReST treatment when delivered four times per week via telehealth, and warrants further large-scale investigation.

Keywords: therapy, intervention, prosody, dyspraxia, video conferencing, Adobe Connect, telespeech, telepractice.

What this paper adds?

What is already known on the subject?

Telehealth is being increasingly used for assessment and treatment of communication disorders. This service delivery method has demonstrated effectiveness for the treatment of many communication disorders, including articulation and phonology impairments, but there is no information about the efficacy of telehealth treatments for CAS. ReST treatment is efficacious for CAS when delivered face to face, producing the acquisition of new pseudo-words, generalization to untreated skills and retention of skills following treatment. This phase 1 study investigates the efficacy of ReST treatment for CAS when delivered by telehealth.

What this paper adds?

Preliminary support for the use of ReST treatment, when delivered four times per week via video conferencing. The results justify larger scale studies of this service delivery method.

Introduction

Children with childhood apraxia of speech (CAS) have difficulty planning and programming the movements required for the production of accurate speech sounds and prosody. Their speech is often characterized by inconsistent errors, inappropriate prosody and disrupted coarticulatory transitions (American Speech–Language–Hearing Association 2007). The difficulties associated with their impairment are often persistent (Lewis *et al.* 2004) with potential effects in a range of linguistic and speech-motor domains (American Speech–Language–Hearing Association 2007). It has been argued that children with CAS require more intensive treatments than other speech-sound disorders (Maas *et al.* 2014, Murray *et al.* 2014, Namasivayam *et al.* 2015), and for a longer period (Skinder-Meredith 2001).

Although several different treatments are used for CAS, most have been investigated in case study or case-series designs and have low levels of evidence regarding their effectiveness (Murray *et al.* 2014, Maas *et al.* 2014). Rapid Syllable Transitions (ReST) is a relatively new treatment for CAS that uses pseudo-word targets with varying lexical stress patterns to target simultaneously articulatory accuracy, fluent transitions between syllables and lexical stress. ReST incorporates motor learning principles to facilitate retention and generalization of treated skills. ReST treatment has demonstrated an improvement in treated items (Ballard *et al.* 2010, Thomas *et al.* 2014), generalization of treatment effects to untreated pseudo-words (Ballard *et al.* 2010, Thomas *et al.* 2014), and to connected speech (Staples *et al.* 2008). A randomized controlled trial comparing ReST treatment with the Nuffield Dyspraxia Programme—Third Edition, demonstrated the efficacy of both treatments (Murray *et al.* 2015). Specifically, ReST treatment resulted in significant acquisition of treated pseudo-words, significant generalization of treatment effects to untreated pseudo-words and real words, and maintenance of treatment effects for 4 months post-treatment (Murray *et al.* 2015). Although typically delivered across four 1-h sessions per week for 3 weeks, ReST is also efficacious when provided across two 1-h sessions per week for 6 weeks (Thomas *et al.* 2014).

Even though effective treatments exist for CAS, many families are unable to access speech pathologists to provide the required treatment, and when treatment is received it is often less frequent and for a shorter duration than necessary (Ruggero *et al.* 2012). These access difficulties are compounded for people who need to see a specialist clinician or who live in rural and remote areas (O’Callaghan *et al.* 2005). Telehealth, with its provision of therapy services at a distance, can improve access to both high-intensity speech-pathology treatments (Mashima and Doarn 2008) and specialist clinicians.

When provided in the client’s home, telehealth eliminates the travel time associated with face-to-face therapy (Reynolds *et al.* 2009), and improves generalization (Theodoros 2013). Telehealth is well accepted by families (Constantinescu 2012) and in some cases is preferable for clients over face-to-face delivery (Ciccia *et al.* 2011). Although the term ‘telehealth’ covers all types of services mediated by technology, the focus of this article is video conferencing, which provides real-time transmission of both audio and visual information.

There is growing evidence supporting the use of video conferencing for speech pathology (for reviews, see Theodoros 2011 and Mashima and Doarn 2008). The effectiveness of video conferencing has been more widely investigated for assessments than for therapy. Video conferencing assessments produce equivalent results to face-to-face assessments in several speech and language areas, including paediatric speech-sound disorders (Eriks-Brophy *et al.* 2008, Waite *et al.* 2012). Despite the promising results from speech-pathology assessment of speech-sound disorders using video conferencing, poor inter-rater reliability has been shown between face-to-face and telehealth assessments for the identification of the presence or absence of voicing, accuracy of fricative phoneme perception, identification of phonemes without visible articulation (e.g., /tʃ/ and /l/) (Eriks-Brophy *et al.* 2008, Waite *et al.* 2006), and perception of abnormal nasal resonance in speech (Hill *et al.* 2006).

Video conferencing as a service delivery model is showing promising results for speech-pathology treatments, particularly treatments that are operationally defined. Effective treatment via video conferencing has been demonstrated for the Lidcombe Program for stuttering (O’Brian *et al.* 2014), the Camperdown Program for stuttering (Carey *et al.* 2014), and the Lee Silverman Voice Treatment (LSVT[®]) for patients with Parkinson’s disease (Constantinescu *et al.* 2011).

Articulation impairments have been effectively treated via video conferencing. In a series of studies culminating in a randomized controlled trial, traditional articulation therapy was shown to be as effective via video conferencing as face-to-face delivery (Grogan-Johnson *et al.* 2013). The participants in Grogan-Johnson *et al.*’s (2013) study had articulation and phonological disorders rather than CAS (S. Grogan-Johnson, personal communication, 6 February 2015) and therefore these findings cannot necessarily be applied to children with CAS. Effective treatments for CAS often focus on prosody or speech movements (Maas *et al.* 2014, Murray *et al.* 2014) rather than targeting specific sound errors in a step-by-step progression.

There is currently no evidence for efficacy of video conferencing for CAS treatments. The compromised sound signal sometimes associated with video conferencing (Keck and Doarn 2014) may potentially

reduce the effectiveness of treatment. Given that speech pathologists have an ethical responsibility to ensure their treatments are effective and efficient (Speech Pathology Australia n.d.), it is important to investigate the efficacy of telehealth for delivering treatment for this population.

In this study we investigated the efficacy of ReST treatment for CAS via video conferencing, with the participants receiving treatment at home, using their own computers and existing Internet connection.

The hypotheses were as follows:

- ReST treatment, delivered four times a week for 3 weeks via video conferencing, will result in:
 - acquisition of targeted speech behaviours, namely accurate production of phonemes, lexical stress pattern and smooth transitions between syllables, in imitated pseudo-words, as perceived by the probe assessor;
 - generalization of this treatment effect to untreated but related imitated speech behaviours:
 - pseudo-words with the same phonemes and lexical stress patterns as treated items;
 - real words with the same number of syllables as the treated items.
 - maintenance of speech gains up to 4 months post-treatment.
- Telehealth treatment will be viewed as comparable or more desirable than intensive face-to-face clinic treatment, as measured via telephone interview with one caregiver per child, 4 weeks post-treatment.

Method

Participants

Eleven monolingual Australian English-speaking children consented to participate in the study. Six children were excluded from the study following assessment, as they did not meet the inclusion criteria defined below. Five children with a diagnosis of CAS aged 5:5 (years; months) to 11:2 completed the study.

Inclusion criteria were (1) consensus diagnosis of CAS (see below), (2) passed pure tone audiometry at 20 dB at 500, 1, 2 and 4 kHz, (3) normal receptive vocabulary (Peabody Picture Vocabulary Test—4th Edition; Dunn and Dunn 2007), and (4) normal oral structure (Oral and Speech Motor Protocol; Robbins and Klee 1987). The diagnosis of CAS was made independently by the first two authors based on the perception of the presence of core perceptual features of CAS (American Speech–Language–Hearing Association 2007) during a battery of speech production tests. There are currently no specific tests or agreed cut-off points for determining the presence of the core perceptual features (American Speech–Language–Hearing Association

2007). We chose relatively low cut-off points for each feature, as we were recruiting children up to 12 years of age and the frequency and/or severity of behaviours associated with the core perceptual features may possibly reduce as children get older. Diagnosis of CAS was given when (1) children < 11 years showed > 40% inconsistency in word production on repeated attempts during the Inconsistency subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd *et al.* 2006) or children aged \geq 11 years showed > 30% inconsistency¹ over three separate administrations of 25 words from the Test of Polysyllables (Gozzard *et al.* 2006); (2) a minimum of 10 words exhibited syllable segregation within words during the Test of Polysyllables (Gozzard *et al.* 2006), indicating difficulty transitioning between syllables; and (3) a minimum of 15% stress pattern mismatches were produced on the Test of Polysyllables, and the examiners perceived abnormal prosody during conversational speech.

Two additional tests were used to provide more detail on the severity of the children's overall language and articulation skills relative to age-matched peers, but were not used to determine suitability for the study: (1) the Clinical Evaluation of Language Fundamentals—Preschool Second Edition (CELF-P2; Wiig *et al.* 2004) or the 4th Edition Australian version (CELF-4; Semel *et al.* 2006), depending on age; and (2) the Goldman–Fristoe Test of Articulation—2 (GFTA-2; Goldman and Fristoe 2000).

The children were assigned pseudonyms. Their performance on the above speech and language tests is reported in table 1.

All children had previously received speech therapy, but did not have any other speech treatment from the start of baseline testing until 1 month post-treatment. During the period between 1 and 4 months post-treatment, none of the participants received speech-sound intervention; however, Emily received therapy to improve her receptive and expressive language skills. The research project was approved by The University of Sydney Human Ethics Committee (reference number 2014/080).

Design

A multiple baseline across participants design (Kazdin 2011) was used in this study. Participants were allocated either three, four, five or six twice-weekly baseline sessions. The treatment commenced after different numbers of baseline sessions to demonstrate that change occurred following the commencement of treatment, rather than after a certain number of baseline sessions. During the treatment phase, each participant's performance was monitored three times; immediately prior to treatment sessions five and nine, and 1 day

Table 1. Participants' initial assessment results

Test	Oliver (5 years; 6 months)	Jack (11 years; 0 months)	Emily (11 years; 2 months)	Luke (5 years; 3 months)	Lachlan (7 years; 6 months)
Clinical Evaluation of Language Fundamentals—Preschool Second Edition (CELF-P2) or Clinical Evaluation of Language Fundamentals—Fourth Edition (CELF 4)^a					
<i>Receptive Language Index</i>					
Standard score	79	106	88	84	75
Percentile rank	8	66	21	14	5
Interpretation	< NL mild	WNL	WNL	< NL mild	< NL moderate
<i>Expressive Language Index</i>					
Standard score	92	112	63	59	70
Percentile rank	30	79	1	0.3	2
Interpretation	WNL	WNL	< NL severe	< NL severe	< NL severe
<i>Peabody Picture Vocabulary Test</i>					
Standard score	90	90	99	88	108
Percentile rank	25	25	47	21	70
Interpretation	WNL	WNL	WNL	WNL	WNL
<i>Goldman–Fristoe Test of Articulation</i>					
Standard score	79	45	75	58	69
Percentile rank	11	<1	<1	2	5
Interpretation	< NL mild	< NL severe	< NL severe	< NL severe	< NL moderate
<i>Test of Auditory Perception—Third Edition, Word Discrimination subtest</i>					
Scaled score	9	9	12	7	10
Percentile rank	37	37	75	16	50
Interpretation	WNL	WNL	WNL	WNL	WNL
<i>Inconsistency Assessment^b</i>					
% Inconsistency	68	32	44	64	48
Interpretation	Inconsistent	Inconsistent	Inconsistent	Inconsistent	Inconsistent
<i>Test of Polysyllables</i>					
% Consonants correct	76	84	85	36	67
% Vowels correct	67	91	88	50	74
% Phonemes correct	73	87	86	42	70
% Stress pattern errors ^c	46	26	32	77	47
% Syllable segregations ^d	20	20	25	21	22
<i>Oral and motor speech protocol</i>					
Structure					
Raw score	23	23	25	24	23
Interpretation	WNL	^	^	WNL	^
Function					
Raw score	94	97	102	107	104
Interpretation	< NL	^	^	< NL	^
Observations	Difficulty coordinating lip and tongue movements in non-speech and speech tasks	Reduced speaking volume, intermittent hypernasality	Inconsistent hypernasality. Loud speaking volume	Difficulty coordinating lip and tongue movements in non-speech and speech tasks	Difficulty imitating multisyllabic words. Incoordination during DDK tasks

Notes: ^aChildren aged 5 years completed the Clinical Evaluation of Language Fundamentals—Preschool Second Edition (CELF-P2), those aged more than 6 years completed the Clinical Evaluation of Language Fundamentals—Fourth Edition (CELF 4); see the participants section for test references; WNL, within normal limits; NL, normal limits; DDK, Diadochokinesis.

^bChildren less than 11 years completed the Inconsistency subset of the Diagnostic Evaluation of Articulation and Phonology (Dodd *et al.* 2006) and those \geq 11 years completed three productions of 25 words from the Test of Polysyllables (Gozzard *et al.* 2006). Inconsistent; see the participants section for further information regarding inconsistency assessment.

^cCalculated using Profile of Phonology (PROPH) software.

^dPercentage of words with at least one perceptually identified absence of smooth joining of the syllables; ^ = outside of age range for normative scores.

post-treatment. Each participant's performance was also monitored three times in the follow-up phase at 1 week, 4 weeks and 4 months post-treatment.

Demonstration of experimental control in multiple-baseline designs is through the replication of the

treatment effect across participants, with staggered introduction of the independent variable across different time points (Kazdin 2011). Although internal validity is typically addressed through replication of the effect, research with children faces a threat to internal

validity as a result of maturation. As an additional safeguard against maturation effects we included a control behaviour to our probe stimuli for each child (see 'Probe stimuli' for details).

Probe stimuli

A 90-item probe list was created for each child to permit analysis of (1) treatment effect, (2) generalization to related, but untreated items, (3) generalization to real words with the same number of syllables as the treated items, and (4) maturational control. The probe stimuli included pseudo-word strings with strong-weak (SW) stress patterns (e.g., /dabəfi/) and weak-strong (WS) patterns (e.g., /kədəfi/). The consonants for the pseudo-word stimuli represented different manner, place and voicing conditions, namely /d/, /k/, /f/ and /b/. The vowels selected for the pseudo-word strings were /a/, /ɔ/, /i/ and /ə/. The probe and treatment stimuli are included in appendix A.

Lachlan, Oliver, Jack and Emily's probe list consisted of 20 SW and 20 WS three-syllable (CVCVCV) pseudo-words, of which 20 (10 SW and 10 WS) were treated and 20 (10 SW and 10 WS) remained untreated, in order to assess generalization to similar but untreated items. The treated items were selected from the set of pseudo-words, and each participant had a different set of treated items. The probe list also included 20 carrier phrases (e.g., I found a -----) with the three-syllable strings to assess generalization effects to sentence level, and 20 three-syllable real words to assess generalization to real words. Additionally, each child had 10 control items, which contained an articulation error or phonological process that we hypothesized would not change during ReST treatment, as it was unrelated to treated items (e.g., a liquid when only plosives and fricatives were trained, or an inter-dental lisp when prosody and nasality were targeted), or it represented a more complex skill level than treated (e.g., clusters). Lachlan's control behaviour was production of word initial /s/ clusters, Oliver and Emily's was articulation of /r/ in initial and medial word position, and Jack's was articulation of /s/ in initial- and final-word position.

Luke's speech difficulties were more severe than the other participants and his treatment stimuli were two syllable pseudo-words. His probe list contained 20 SW and 20 WS two syllable (CVCV) pseudo-words, with 10 SW and 10 WS items randomly selected for treatment, and 10 of each kept to assess generalization to untreated items. His probe list also included 20 three syllable (CVCVCV) pseudo-words, to assess performance on more complex pseudo-words, 10 two syllable real words, and 10 three syllable real words, to assess generalization to real words. Luke's control behaviour was the production of initial /l/ clusters (/pl/, /bl/, /kl/, /fl/ and /gl/).



Figure 1. Microphone and headphone set-up

Equipment

Video conferencing was conducted using Adobe Connect, version 8, which had the function to share documents and interactive workspaces as well as transmit real-time audio and visual information. The speech-pathology clinicians used either a Dell Latitude E6320 laptop computer with an inbuilt web camera or a custom-built Bosch P8C WS desktop computer with Logitech C930e web camera. Clinicians wore a USB headset (Sennheiser PC 8 or Logitech H540). All participants used their home computer, with broadband Internet connection. Participants wore a Sennheiser PC 8 USB headset around the neck with the microphone positioned approximately 10 cm from the mouth to record sound and Yellowstone YSYROHRD headphones over the ears (figure 1). A separate headphone and microphone for participants was used to enable a 3.5-mm audio splitter to connect to the caregiver's Yellowstone YSYROHRD headphones allowing them to hear the child's session. All sessions were recorded through Adobe Connect for later assessment of treatment fidelity, scoring reliability, and for student training purposes. The sessions were also recorded at the participant's home using an Olympus VN-711PC digital voice recorder; however, all data reported here are based on the Adobe Connect recordings.

The face-to-face initial assessments were audio recorded with an AKG C520 headset microphone and Roland Quad Capture UA-55. They were video recorded using a Bosch NBN-832V-P camera, and an Electrovoice RE90HW microphone connected to a Bosch DIVAR IP 7000 2U DVD.

Procedure

The first author, a qualified speech pathologist, carried out the face-to-face eligibility assessments and video conferencing baseline probes. Jack and Oliver were

treated by qualified speech pathologists experienced in ReST treatment; while Lachlan, Luke and Emily were treated by trained speech pathology students, under the supervision of the first and second authors. The same clinician treated Emily and Lachlan. One clinician treated each child for the duration of the treatment phase.²

Baseline and probe sessions

Identical procedures were used for baseline and the probe sessions. The probe list items were presented in one of three randomized orders. The participants viewed a PowerPoint slide show, with the orthography for each pseudo-word item and a picture plus orthography for each real-word item and the sound file of an Australian English female speaker producing each item. As the participant viewed each slide, the parent played the sound file for the item and the child imitated the word. Imitation was used due to the non-familiarity of the pseudo-word items and to ensure consistency of procedure between pseudo- and real-word items. During the PowerPoint slide show, the clinician could see and hear the participant via the web camera and microphone, and the participant could hear the clinician, but see only the PowerPoint slide show.

Technology set-up

Prior to the baseline sessions, each participant had one or two 30-min web-conferencing familiarization sessions where the treating clinician and child talked via video conference, played interactive web-based games, and solved any technical difficulties with equipment or connectivity.

Technology rating

Following each session, the treating clinician completed a form noting any technical issues, whether the issues were resolved and the strategies employed. The clinician also marked a line on a 10-cm visual analogue scale to rate the technology in the session, from 'very poor' to 'excellent'.

Parent satisfaction

Four weeks post-treatment, telephone interviews were conducted with the treating clinicians and the parents. During the semi-structured interview, the parents and clinicians used a 10-point rating scale (e.g., 0 = not convenient at all, 10 = very convenient) to rate the convenience of the sessions, their perception of the child's motivation and their overall satisfaction with the telehealth mode of treatment.

Treatment

The ReST treatment was used, following the procedure described in Murray *et al.* (2012). However, unlike Murray *et al.* (2015), all children in this study imitated the stimulus items, while looking at the written stimulus rather than reading the items. Each session began with approximately 10 min of pre-practice to explain the task and ensure the children had a reference of correctness for the target stimuli. During pre-practice, the participants (1) viewed a card with the written pseudo-word via the webcam, (2) listened and watched the computer monitor while the clinician produced the selected written pseudo-word, from the 20 treatment items, and (3) attempted to imitate the word production.

The participants were provided with knowledge of performance (KP) feedback immediately following each production (e.g., 'That word was broken, the parts were separated. Try to join the parts together smoothly'). A variety of cueing techniques were employed such as breaking the words into syllables and rejoining, representing relative syllable duration with magnetic strips on a whiteboard, slowing overall rate of production, and cueing about correct articulator placement. Once five items were produced correctly with modelling and shaping, the participant moved into the practice phase. The pre-practice phase lasted for up to 25 min in sessions 1, 2 and in any session where a child progressed to a new level of treatment, and approximately 10 min in all other sessions.

In the practice phase, each participant aimed to complete 100 trials ($\bar{x} = 99$, $SD = 9.33$): five trials each of the 20 treated items, in random order. The clinician provided a live model of the item for the child to imitate during the practice trials. Knowledge of results (KR) feedback (i.e., feedback about whether the item was correct or incorrect) was provided on approximately 50% of the items after a delay of 3–5 s. After every 20 trial items, a 2-min rest break was provided.

Once a participant achieved $\geq 80\%$ correct in two consecutive practice sessions, the client began treatment on the next, more complex treatment level (see Murray *et al.* 2012 for levels in ReST treatment). The progression criterion was met by Jack in session 5, and Emily in session 10, and these children moved to treatment on pseudo-words at the end of carrier phrases (e.g., 'She has a big /dɒfabi/' or 'There's a /dɒbɒfi/') from sessions 6 and 11 respectively.

Dependent measures and data analysis

The probe assessors made perceptual judgments about each probe item with regard to the accuracy of the phonemes, stress pattern and fluency of syllable transitions. Judgements were made about each construct

individually and, in order to be counted as correct, the probe item needed (1) correct sounds, (2) correct lexical stress and (3) smooth connection of the syllables. The dependent measure was the percentage of items correct (i.e., with correct sounds, lexical stress and smooth connection between the syllables). The first author conducted all baseline assessments. A rater blinded to the phase of treatment and baseline level of speech skill conducted the probe assessments. Intra- and inter-rater reliability was calculated on 20% of each baseline session, probe assessment and treatment session.

Data for each participant were graphed for visual analysis. Visual analysis consisted of examining the level, trend, variability, overlap and immediacy of effect. Visual analyses were supported with statistical analyses where possible. In order to do so, we tested each child's data for independence by preliminary analyses of variance comparing phases, recording residuals from these analyses and testing the residuals for autocorrelation. With the exception of Lachlan's untreated pseudo-words and carrier phrases, and Emily's real words, in all cases the lag 1 correlation of the residuals was non-significant, indicating no evidence that the assumption of independence was violated in most cases. Where other analysis of variance (ANOVA) assumptions were met, ANOVAs and Helmert planned orthogonal contrasts were performed for each participant to test for differences across phases (baseline, treatment, follow-up) within behaviours (treated pseudo-words, untreated pseudo-words, untreated real words, more complex pseudo-words or pseudo-words in carrier phrases and control words). In each case, the first Helmert contrast compared the average within-participant performance in the baseline phase with average performance over treatment and follow-up phases, and the second contrast compared the average within-participant performance in the treatment phase with the follow-up phase. A study-wide adjustment to the significance level, to account for multiple comparisons, was not performed. This is because the primary method of analysis was visual analysis, as is common in single-case design, with the statistical analyses used to confirm the results of visual analysis. Significance at both 0.05 and 0.01 levels are indicated in table 3, and readers are advised to use caution when interpreting significance values between 0.05 and 0.01. Where data were autocorrelated, only visual analysis was performed.

In order to test for maintenance of treatment effect within the follow-up phase, post-hoc planned orthogonal contrasts were performed at the data points within the follow-up phase, with the participants' data pooled. Contrasts were conducted of average performance across participants at (1) 1 day post-treatment with later points (i.e., 1 week, 1 month and 4 months post-treatment combined), (2) 1 week post-treatment

Table 2. Reliability information

	Probe items ^a			
	Pseudo-words	Real words	Control sounds	Treatment items ^a
<i>Judgements of correctness</i>				
Intra-rater	92	91.9	93.5	91
Inter-rater	89	87.3	81.5	88
Broad phonemic transcription				
Inter-rater	89.4	82.5	92.8	95
Inter-rater	84.9	78.5	80.5	94

Note: ^aPercentage agreement.

with later points (i.e., 1 and 4 months post-treatment combined), and (3) 1 month post-treatment with 4 months post-treatment. Effect sizes were calculated using the protocol described by Beeson and Robey (2006): $d_2 = (\text{mean score in follow-up phase} - \text{mean score in baseline phase})/\text{pooled standard deviation}$.

Reliability

Inter- and intra-rater reliability was calculated for phonemic transcription and the scoring of articulation accuracy, stress pattern and fluency of syllable transitions. Given the indications in the literature that perception of some sounds via video conferencing can be unsatisfactory (see the Introduction for details), reliability was calculated separately for pseudo-words items, real-word items and control items (table 2).

Treatment fidelity

The first author examined a randomly selected 10 min of each session for treatment fidelity. Assessment was made of the accuracy of the clinician's model, the number of trials given feedback, the accuracy of the feedback, the type of feedback (i.e., KP in pre-practice and KR in practice), and the timing of feedback. Average fidelity for treatment sessions was 95% (SD = 6.1, range = 75–100). Fidelity was lowest in the first two sessions, involving clinicians giving feedback without sufficient delay, and giving KP rather than KR feedback in the practice phase.

Results

Effects of treatment

Oliver's per cent accuracy with the to-be-treated items during baseline was 0–10% (figure 2, panel A). His per cent accuracy steadily improved during the treatment phase to 70%, and the difference between the baseline phase and the later phases was significant. The results of all significance testing can be found in table 3.

Table 3. Planned contrasts and effect sizes

	Word set	Effect size $d_2 =$	BL versus later (i.e., T and FU combined)		T versus FU		Change
			$t=$	$p=$	$t=$	$p=$	
Oliver	Treated pseudo-words	9.64	6.595	0.001**	0.835	0.437	–
	Treated pseudo-words in phrases	2.04	2.418	0.065	5.156	0.68	–
	Untreated pseudo-words	1.79	3.397	0.015*	1.343	0.229	–
	Untreated real words	5.73	6.711	0.001**	0.731	0.497	–
	Control /r/ articulation	1.63	1.277	0.251	0.583	0.583	–
Jack	Treated pseudo-words	3.59	8.333	< 0.001**	0.177	0.111	–
	Treated pseudo-words in phrases	2.30	3.878	0.004**	0.282	0.784	–
	Untreated pseudo-words	3.59	8.333	< 0.001**	0.176	0.111	–
	Untreated real words	6.34	9.929	< 0.001**	1.136	0.286	–
	Control /s/ articulation	0.00	1.065	0.316	1.523	0.168	–
Emily	Treated pseudo-words	4.65	10.121	< 0.001**	2.705	0.003**	↓
	Treated pseudo-words in phrases	2.00	3.16	0.013*	0.321	0.757	–
	Untreated pseudo-words	3.48	5.172	0.001**	4.84	0.525	–
	Untreated real words	0.70	0.75	0.473	0.357	0.731	–
Luke	Treated pseudo-words	21.24	16.588	< 0.001**	1.452	0.193	–
	Untreated three-syllable pseudo-words	3.20	2.273	0.060	1.179	0.634	–
	Untreated pseudo-words	13.16	17.358	< 0.001**	0.515	0.275	–
	Untreated real words	3.12	4.37	0.003**	1.029	0.341	–
Lachlan	Control /l/ clusters	0.64	5.952	0.001**	11.111	< 0.001**	↓
	Treated pseudo-words	6.79	3.791	0.009**	1.329	0.232	–
	Treated pseudo-words in phrases						
	Untreated pseudo-words						
	Untreated real words	3.87	4.6	0.004**	2.741	0.034*	↑
	Control /s/ clusters	0.07	0.532	0.612	0.931	0.390	–

Note: Effect size = Cohen's d_2 using pooled standard deviations (Beeson and Robey 2006); BL, baseline phase; T, treatment phase; FU, follow-up phase; **significant at 0.01; *significant at 0.05; –, No difference between treatment and maintenance phase; ↓, decrease in the follow-up phase; ↑, increase in the follow-up phase. Contrasts for Emily's untreated real words and Lachlan's pseudo-words in phrases and untreated pseudo-words were not calculated because the data showed autocorrelation.

Jack's per cent accuracy during the baseline phase with the to-be-treated items ranged from 30% to 35% (figure 3, panel A). During the treatment phase, his per cent accuracy increased to 85–95%, resulting in a significant difference between baseline performance and later phase performance. Jack reached the a priori criterion of 80% accuracy on treated behaviours over two consecutive treatment sessions in the fifth treatment session. His therapy target was therefore changed from single pseudo-words to pseudo-words in carrier phrases from session 6. During baseline, Jack's per cent accuracy on treated pseudo-words in carrier phrases ranged between 0% and 10% (figure 3, panel B). In probe 7, following the introduction of treatment on single pseudo-words, his performance on pseudo-words in carrier phrases improved to 90% accuracy, suggesting generalization of treatment effects (see below). His accuracy with carrier phrases in probes 8 and 9 was similar to probe 7.

Emily's performance with the to-be-treated items during the baseline phase ranged from 45% to 60% accuracy (figure 4, panel A). During the treatment phase, her treated pseudo-word accuracy ranged from 84% to 90%, and planned contrasts confirmed Emily had significantly better performance in later phases than in baseline. Emily reached the a priori criterion of 80% accuracy on treated single pseudo-words over two consecutive treatment sessions in the 10th treatment session. Her treatment goal changed to the production of pseudo-words in carrier phrases from session 11. Figure 4, panel B, shows that during baseline Emily's per cent accuracy on treated pseudo-words in carrier phrases ranged between 5% and 15%. Her accuracy with pseudo-words in carrier phrases improved when she started treatment on single pseudo-words, suggesting generalization of treatment effects to more complex stimuli (see below).

Luke's per cent accuracy with the to-be-treated items during the baseline phase ranged from 5% to

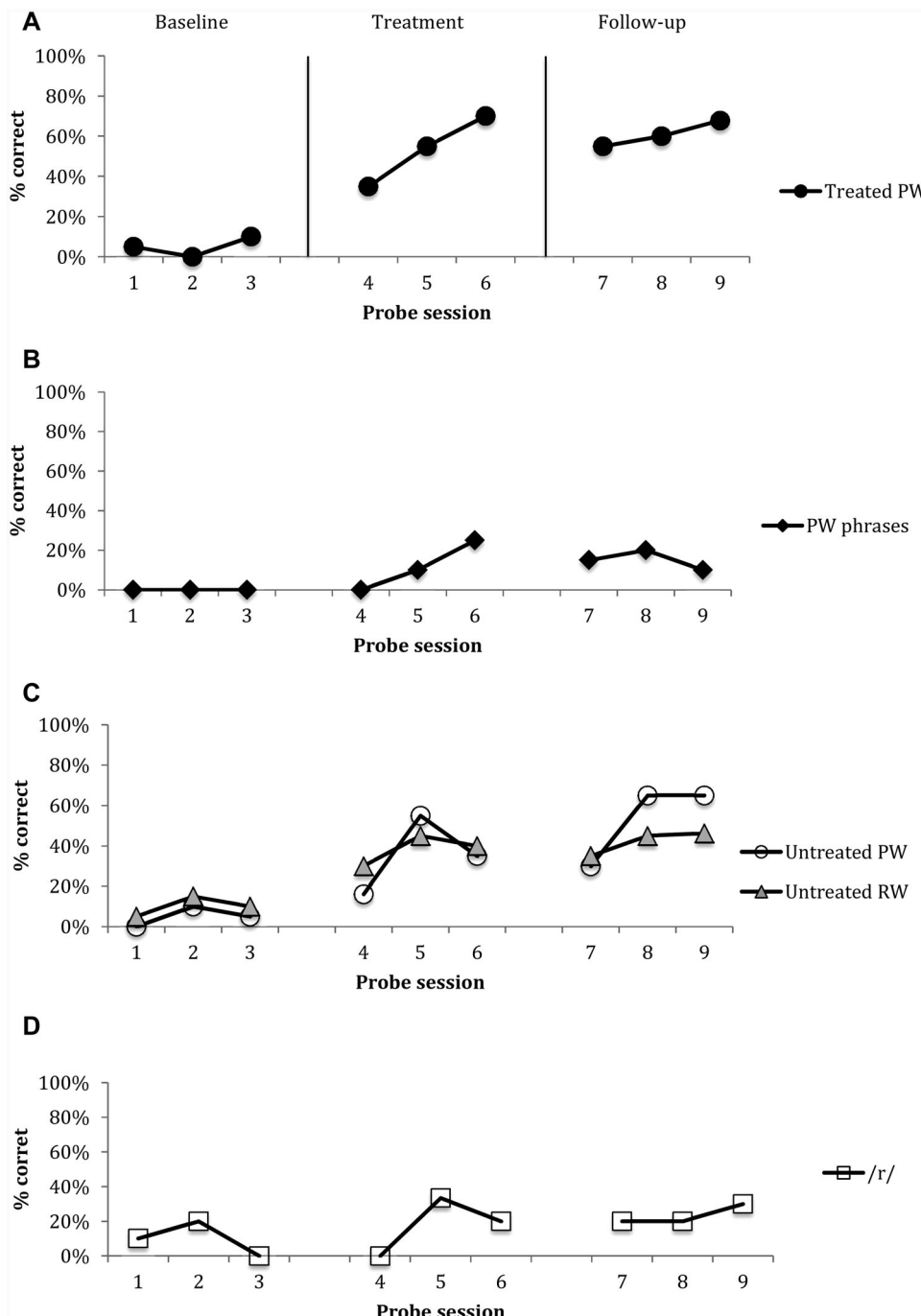


Figure 2. Oliver's results. PW, pseudo words; RW, real words.

10% (figure 5, panel A). During the treatment phase his accuracy with treated pseudo-words was 50–65% and planned contrasts confirmed that the improvement from baseline to the later phases (treatment and follow-up) was significant.

Lachlan's per cent accuracy with the to-be-treated items during baseline was 0–11% (figure 6, panel A). Within the treatment phase his per cent accuracy

steadily improved to 75%, resulting in a significant difference between the baseline phase and the later phases.

Generalization of treatment effects

Oliver showed significant generalization to untreated pseudo-words and untreated real words (figure 2,

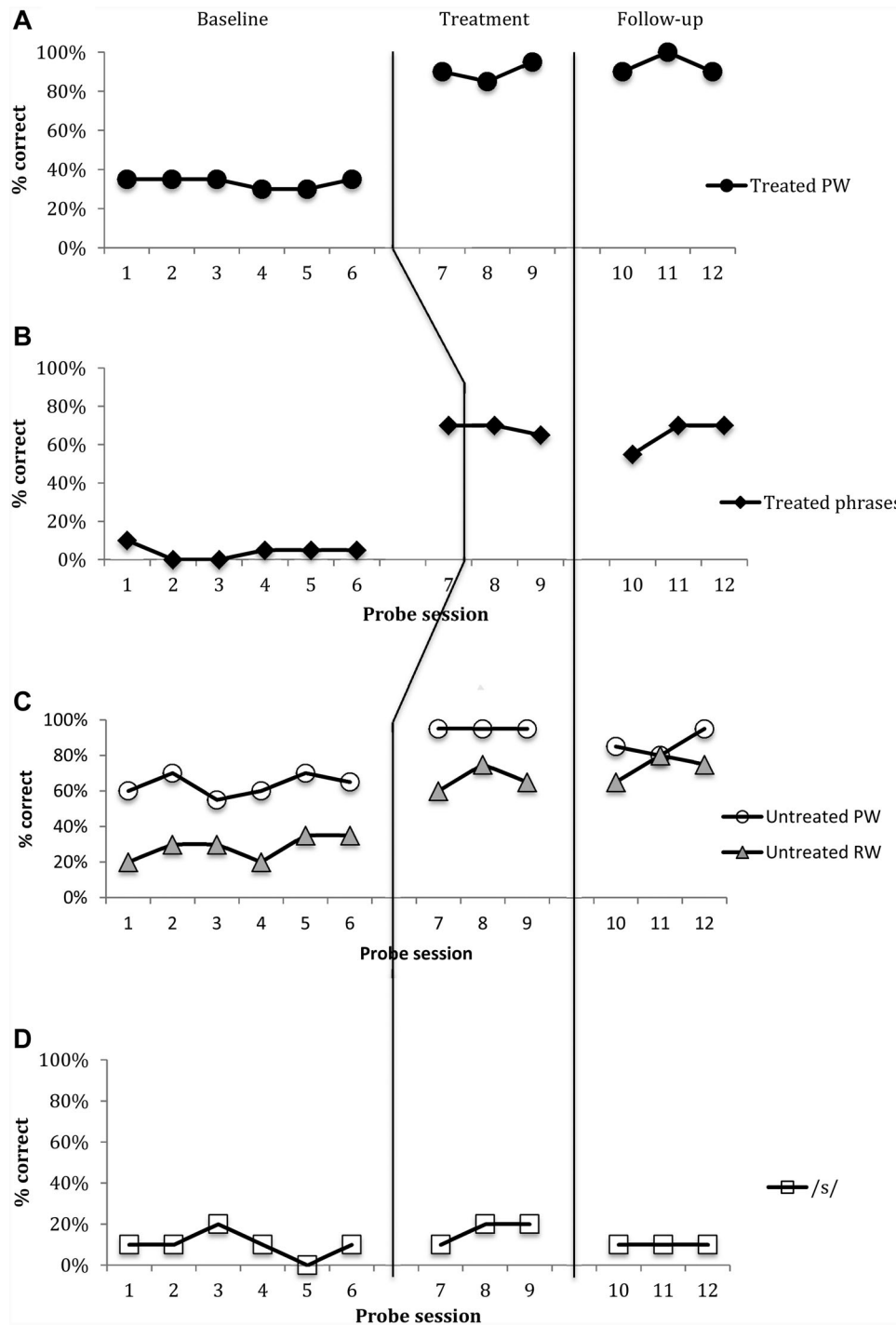


Figure 3. Jack's results. PW, pseudo words; RW, real words.

panel C). During baseline, his per cent accuracy with untreated pseudo-words and untreated real words was 5–15% and 0–10% respectively. During the treatment phase, his accuracy for these items improved significantly to 16–55% and 30–45% respectively. Visual inspection of Oliver's accuracy with pseudo-words in carrier phrases (figure 2, panel B) indicates a small

improvement in these items during the treatment phase, which was not statistically significant.

Jack generalized his skill to similar, but untreated, pseudo-words and untreated real words, as shown in figure 3, panel C. In baseline, his accuracy was 60–70% for untreated pseudo-words and 20–35% for untreated real words. During the treatment phase, his performance

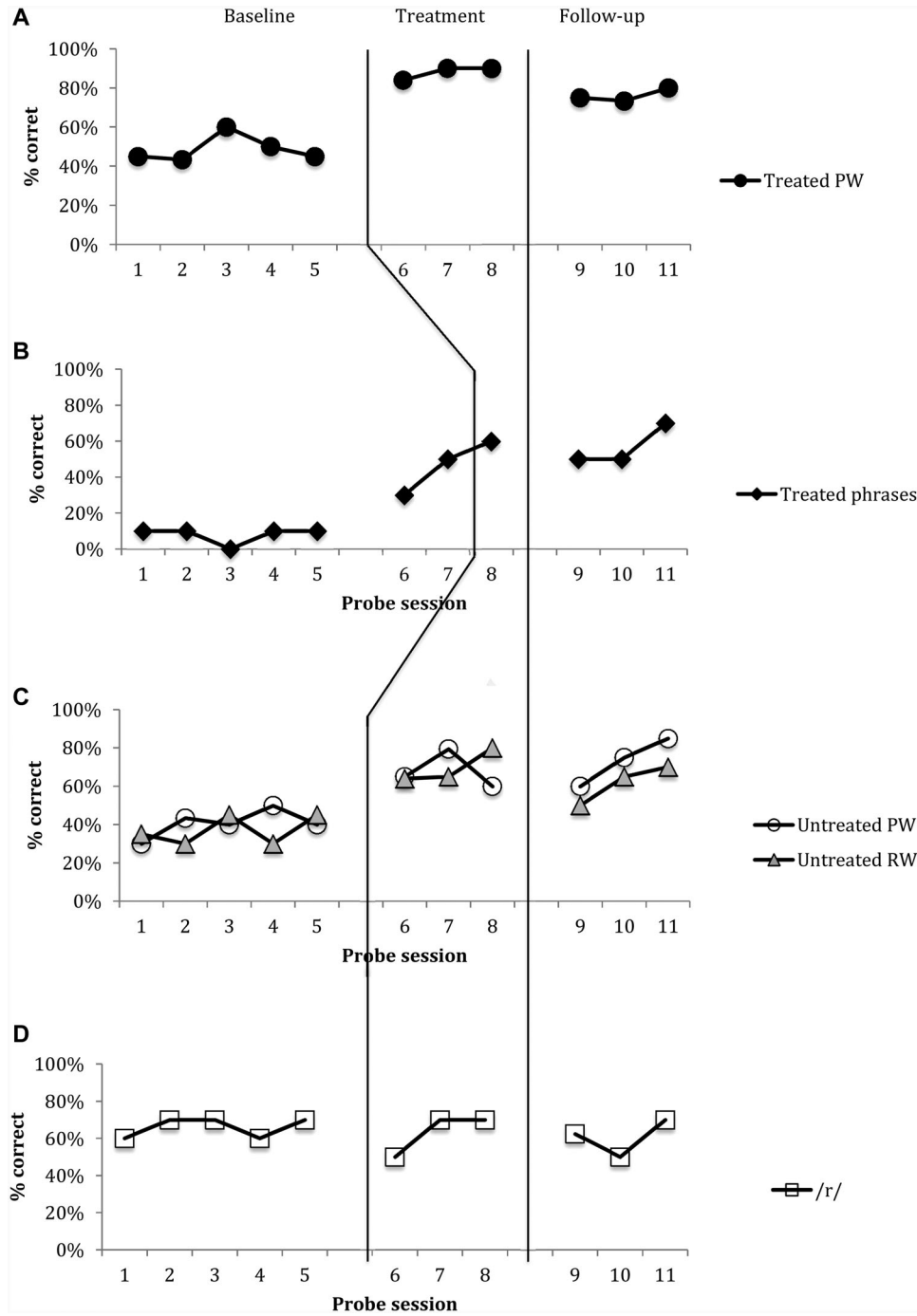


Figure 4. Emily's results. PW, pseudo words; RW, real words.

improved to 95% accuracy for untreated pseudo-words and 60–75% for untreated real words. Planned contrasts confirmed these differences were significant. As treatment shifted to treated pseudo-words in carrier phrases after probe 7, Jack's performance with treated words in carrier phrases in probe 7 was compared with his performance in the other baseline probes (figure 3, panel B). Jack's accuracy with pseudo-words in carrier

phrases increased from < 10% in probes 1–6 to 70% in probe 7 following the introduction of treatment on single pseudo-words. Visual inspection indicated that there was no difference between performance on treated pseudo-words in carrier phrases between probe session 7 and later probe sessions, suggesting generalization to treated carrier phrases occurred once treatment began on the pseudo-words (figure 3, panel B).

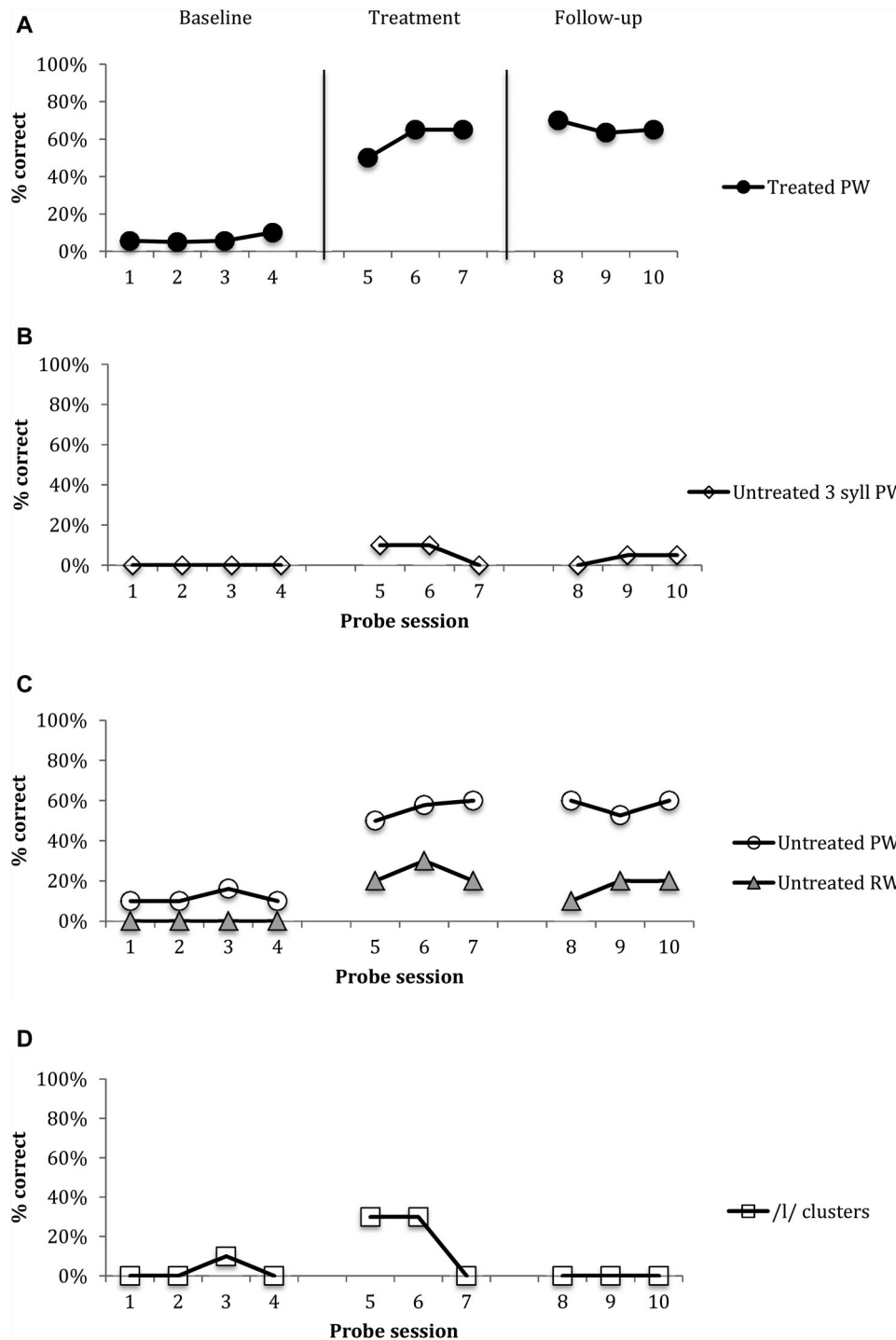


Figure 5. Luke's results. PW, pseudo words; RW, real words; syll., syllable.

Emily showed generalization to untreated pseudo-words and untreated real words (figure 4, panel C). During baseline, her per cent accuracy with untreated pseudo-words and untreated real words was 35–50% and 35–45% respectively. During the treatment phase, her accuracy for these items improved to 60–79% and 65–80% respectively, and the difference between

performance in the baseline phase and later phases was significant. As discussed previously, Emily also showed generalization to carrier phrases with pseudo-words prior to treatment at the carrier phrase level. We compared her accuracy with carrier phrases in probe 7 (the last probe prior to treatment on pseudo-words in phrases) to probes 1–5 (prior to treatment on single

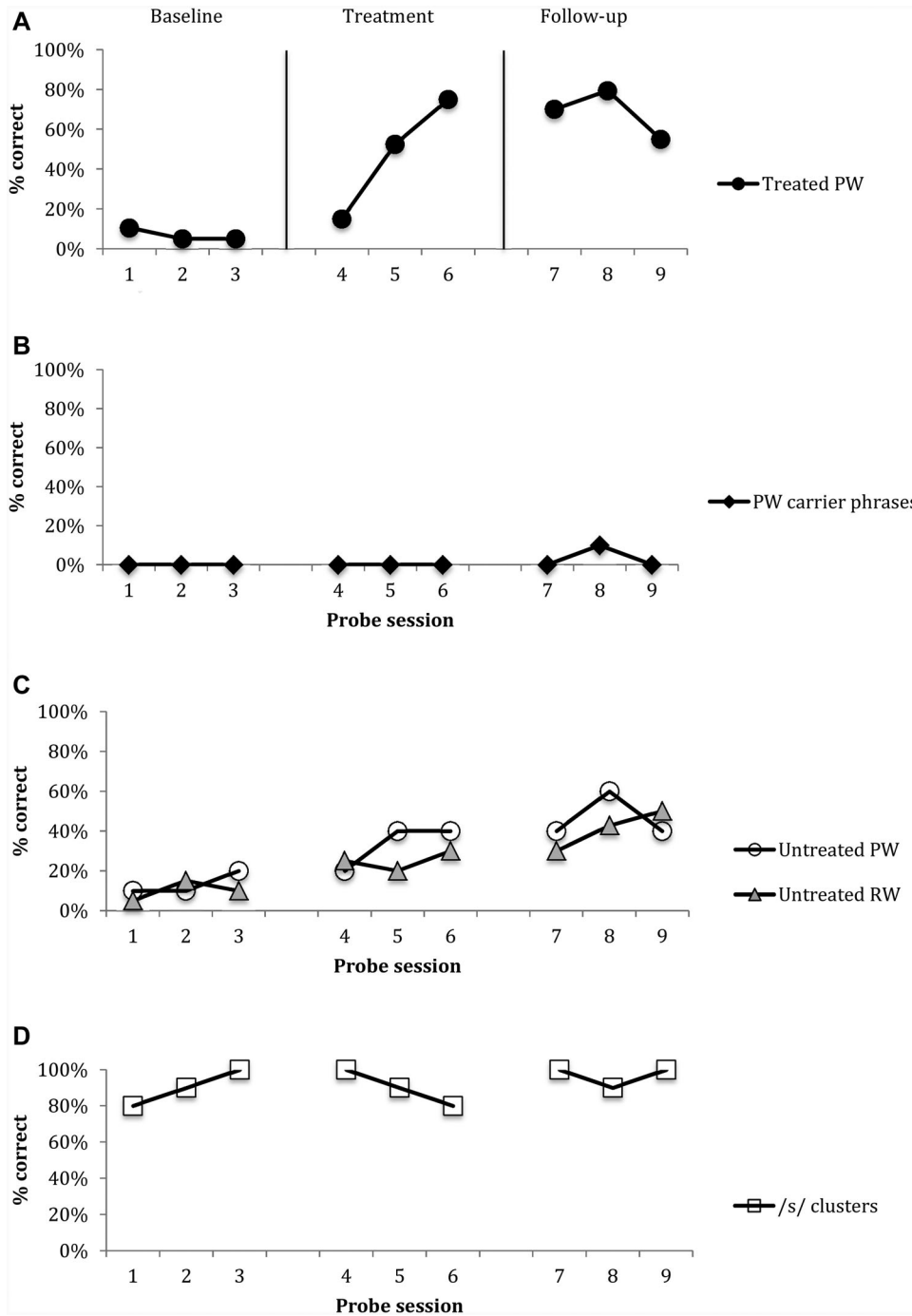


Figure 6. Lachlan's results. PW, pseudo words; RW, real words.

pseudo-words). In the first five probe sessions Emily achieved < 10% accuracy on treated pseudo-words in carrier phrases. Her accuracy with this behaviour improved steadily once treatment began on single pseudo-words, resulting in 50% accuracy in probe 7. This suggests generalization to carrier phrases with pseudo-words once treatment began on single pseudo-words (figure 4, panel B).

Luke generalized his skills to similar, but untreated, pseudo-words and untreated real words, as shown in figure 5, panel C. During baseline, his accuracy with untreated pseudo-words and untreated real words was 10–16% and 0% respectively. His accuracy improved on these untreated items during the treatment phase resulting in accuracy levels of 50–60% for untreated pseudo-words and 20–30% for untreated real words.

Planned contrasts confirmed that these improvements were significant. Although visual inspection indicates a small, temporary improvement with more complex items (three-syllable pseudo-words; figure 5, panel B), the change in these items between the baseline phase and later phases (i.e., treatment and follow-up) was not significant.

Lachlan showed significant generalization to untreated pseudo-words and untreated real words (figure 6, panel C). During baseline, his per cent accuracy with untreated pseudo-words and untreated real words was 10–20% and 5–15% respectively, both with slightly rising baselines. During the treatment phase, his accuracy with these items improved to 20–40% and 20–30% respectively with the slope greater than predicted by the rising baseline. His untreated pseudo-words showed autocorrelation of the residuals at lag 1 prohibiting statistical analyses. Planned contrasts indicated a significant difference between baseline and later phase performance on untreated real words. Visual inspection indicates Lachlan did not show generalization to pseudo-words in carrier phrases (figure 6, panel B); statistical analysis was not conducted on this data set due to autocorrelation of the residuals.

Maintenance of treatment and generalization effects

Most of the participants' treatment and generalization gains were maintained for 4 months post-treatment. Oliver, Jack, Lachlan and Luke maintained all treatment and generalization effects throughout the follow-up period. They had higher per cent accuracy at all follow-up points than baseline levels, for each of treated pseudo-words, similar but untreated words, and untreated real words. Planned contrasts revealed no significant difference between treatment phase and follow-up phase accuracy for any of these items for Oliver, Jack and Luke, supporting maintenance of effects to 4 months post-treatment. Lachlan had significantly higher accuracy in the follow-up phase than the treatment phase for untreated real words, indicating improving performance following the withdrawal of treatment. Jack also maintained his skill with treated pseudo-words in phrases. His per cent accuracy at two of the follow-up points was at the same level as probe 7 (the final probe prior to treatment on carrier phrases), and at all follow-up points was higher than baseline levels. No significant difference was found between his treatment phase and follow-up phase performance on pseudo-words in carrier phrases, supporting maintenance of skill.

Emily maintained some of her treatment gains and all of her generalization gains. With regard to maintenance of treatment gain, Emily lost some of her gain with treated pseudo-words. She had significantly lower accuracy in the follow-up phase for these items than the

treatment phase, even though all follow-up points had higher accuracy than baseline levels. She did however maintain her treatment gain with treated pseudo-words in phrases. For these items, two follow-up points had the same per cent accuracy as probe 7 (the final probe prior to treatment on those items), and one had higher accuracy. Her performance was above baseline levels at all follow-up points for untreated pseudo-words, and untreated real words, and there was no significant difference between the treatment and follow-up phase accuracy these items, indicating maintenance of generalization effects.

In order to monitor the participants' progress at different time points *within* the follow-up phase, the data for the four participants were grouped and Helmert planned orthogonal contrasts were performed. There was no significant difference between the participants' performance at any of the time points, indicating stable, rather than improving or deteriorating performance in the follow-up phase'.

Control behaviour

Oliver, Jack and Emily did not show significant change in the behaviours we selected to monitor for maturational control (/r/, /s/, /r/ respectively) between the baseline and later phases. Luke's accuracy with the behaviour we selected to monitor to control for maturation effects (/l/ clusters), significantly improved during the treatment phase, and then significantly decreased in the follow-up phase. With regard to Lachlan, the behaviour we selected to monitor to control for maturation effects (/s/ clusters), demonstrated a ceiling effect (80–100% correct) in the baseline phase, prohibiting adequate evaluation of change during the treatment phase. However Lachlan's performance on a stimulus generalization measure (production of treated pseudo-words in carrier phrases) showed no significant change during the entire research period.

Adequacy of technology

Although 61% of the sessions were rated by the treating clinician as having technology difficulties, only one of the 113 sessions (< 1%) was cancelled due to a technical issue, namely the family had exceeded their service provider's monthly data allowance. One additional session was conducted partly by telephone, due to issues with sound transmission during the video conference. At the time of the final follow-up appointment, we assessed the speed of connection for all participants and clinicians. The download speed was above 50 Mbps for Jack, Emily and Lachlan, and below 4 Mbps for Oliver and Luke. Oliver and Luke had lower clinician ratings of technological adequacy than the other participants, with

average ratings of 5.45 and 6.73 out of 10 respectively, compared with an average rating for the other children across all sessions of 8.40. The most frequent technical difficulties experienced were difficulty establishing audio connection, web-camera freezing, and latency in the audio signal. At technology adequacy ratings of less than four (9% of sessions), clinicians reported feeling frustrated, annoyed, stressed, and disappointed with the technology. At technology adequacy rating levels above four (91% of sessions), clinicians reported feeling 'fine', 'comfortable', 'OK' and 'great'.

Satisfaction with video conferencing

The parents were very satisfied with the video conferencing treatment (average score = 9.5, range 7.5–10), and they reported their children were motivated to participate in video conferencing sessions (average score = 8, range = 6.5–10) and they found the home-based treatment very convenient (average score = 9.7, range = 8.5–10). The treating clinicians reported high levels of satisfaction (average score = 8.75, range 7.5–10) and convenience (average score = 9.25, range = 8.5–10) with the telehealth treatment.

Discussion

This study aimed to evaluate the efficacy of ReST treatment for children with CAS when provided by video conferencing. We hypothesized that treatment via video conferencing would result in (1) significant improvement in imitated pseudo-words, (2) significant generalization to related but untreated imitated speech behaviours, and (3) maintenance of treatment and generalization effects. The hypotheses were supported with all five children showing positive gains, and four of the five children maintaining their gains to 4 months post-treatment.

Experimental control was indicated by the establishment of stable baselines prior to the introduction of treatment, and the demonstration of improved performance on the dependent variable when treatment commenced for all five children. Additionally, control for maturation was demonstrated for all five children. Three children (Oliver, Jack and Emily) made no significant change with the behaviour we selected as a maturational control. For Luke, the behaviour we selected for this purpose, (/l/ clusters), co-varied with the treatment. His return to baseline levels following the withdrawal of treatment argues against a maturation effect. For Lachlan, the behaviour we selected to monitor for signs of maturation (/s/ clusters), demonstrated a ceiling effect in the baseline phase. Although a behaviour with lower levels of baseline performance would have ideally been selected, Lachlan did not have another speech behaviour appropriate for this purpose. His perfor-

mance on a stimulus generalization task (production of pseudo-words in carrier phrases) was stable throughout the research period. Although unrelated behaviours are usually selected to monitor for maturational change, an alternative way is to monitor for stimulus generalization. Lachlan's lack of change with a stimulus generalization task argues against maturational change, and supports internal validity.

Video conferencing ReST treatment had similar effects to face-to-face treatment (Ballard *et al.* 2010, Murray *et al.* 2015, Thomas *et al.* 2014). Both service delivery methods resulted in significant acquisition of pseudo-words, with large effect sizes. Significant generalization to untreated but related behaviours, and maintenance of treatment and generalization gains to 4 months post-treatment was shown in both the face-to-face and telehealth modality.

Two of the participants not only generalized to untreated items at the same level as treatment, but also to more complex behaviours. Emily and Jack, who generalized to the more complex behaviour of pseudo-words in carrier phrases, had milder speech difficulties initially than the other participants, were older, had accuracy levels above 80% during treatment and some minimal knowledge of the more complex behaviour in baseline. Greater generalization in ReST treatment has been previously demonstrated for children with milder speech difficulties (Ballard *et al.* 2010, Thomas *et al.* 2014) and ReST treatment is generally more effective for older children with milder speech difficulties (Murray *et al.* 2013). Given that generalization to more complex behaviours occurred prior to treatment at that level, it raises the question of whether the children required treatment on the more complex behaviour. Further investigation of generalization to more complex behaviours during ReST treatment is warranted.

With the exception of Emily's accuracy with treated pseudo-words, all children maintained their gains to 4 months post-treatment. Emily's loss of some treatment gain with single pseudo-words is difficult to explain, particularly as she had high levels of treatment accuracy and strong generalization. Perhaps she did not maintain sufficient focus on the single pseudo-words after her treatment moved to phrases. Like the other participants, on all other behaviours, Emily had stable performance in the follow-up phase. This stable performance in the follow-up phase was also shown in face-to-face ReST treatment delivered twice weekly (Thomas *et al.* 2014), while face-to-face ReST treatment provided four times weekly resulted in significant ongoing improvement during the follow-up phase (Murray *et al.* 2015). This present study was different to that of Murray and colleagues in two significant ways: children with receptive language impairments were included and the mode of treatment was video conferencing rather than face to

face. Either of these factors, or a combination of the two, may account for the superior performance in the maintenance phase for children receiving face-to-face treatment versus video conferencing treatment of the same intensity.

Three of our participants had receptive language impairments, and four had expressive language impairments. The treatment effect for children with language impairments, particularly receptive impairments, may potentially be reduced. However, given that all participants demonstrated significant acquisition of the targeted pseudo-words and generalization effects, any limitation associated with the inclusion of participants with language impairments is minimal.

The stable performance during maintenance in this study was a positive finding, given the relatively low levels of treatment accuracy shown by Lachlan, Luke and Oliver. Previous studies with ReST and other motor speech disorders have indicated that high levels of treatment accuracy, around 70%, for approximately five treatment sessions are generally required for maintenance of treatment gains (Ballard *et al.* 2010, Wambaugh *et al.* 2013). ReST treatment, with its use of motor learning principles to facilitate generalization and maintenance, has previously demonstrated maintenance of treatment gains, even with treatment accuracy levels below 70% (Staples *et al.* 2008, Thomas *et al.* 2014). These findings suggest that clinicians may be able to use a lower criterion for treatment accuracy than is currently recommended for ReST treatment.

With regard to the technology used in the sessions, although the majority of the sessions had some technical difficulty, fewer than 1% of sessions were cancelled, indicating that the technical issues were tolerable for the families. Audio latency was the most troubling technical issues because it affected the interaction between clinician and client, as well as the ability to provide timely feedback and no solution was available for sessions with audio latency. Although most of the other technical issues could be resolved, in some cases problem solving took up to 10 min, which was more than 15% of the session. Parents reported that the two familiarization sessions were valuable for improving their technical skill and confidence. The time required for solving technical problems and familiarizing families with video conferencing systems needs to be factored in when considering using telehealth treatments.

Despite the technical challenges, ReST treatment was efficacious in this format. It may be that the nature of a high-production trial treatment with minimal need for physical prompts such as ReST is well suited to video conferencing. CAS treatments requiring more hands-on cueing such as Dynamic Temporal and Tactile Cueing (DTTC) (Strand *et al.* 2006) may be less suitable for video conferencing.

Parents and clinicians found the system convenient, motivating for the child, and were satisfied with their experience of therapy via video conferencing. The high levels of satisfaction and convenience may be related to the interactive games played using Adobe Connect's 'draw' function during session breaks and the reduction in travel time with home-based video conferencing. This high satisfaction is in keeping with previous telehealth studies (e.g., Constantinescu 2012). The children attended all of their treatment and probe sessions. It is possible that the benefits in terms of convenience helped outweigh technical difficulties experienced.

The reliability of phonemic transcription was similar in this study to face-to-face ReST treatment (cf. Thomas *et al.* 2014). Based on previous research indicating difficulty perceiving high frequency sounds, clusters, and phonemes without visible articulation (Eriks-Brophy *et al.* 2008, Waite *et al.* 2006) we would not have been surprised to find poor reliability for the control items (*/s/*, */l/* clusters, */s/* clusters, and */r/*), however the average intra- and inter-rater reliability for the control items was acceptable at 93.5% and 81.5% respectively.

Limitations and future directions

This was a small, phase 1 study. It would be beneficial to investigate the use of video conferencing for ReST treatment in a larger group study, and to investigate the factors affecting treatment outcomes for children. It would also be beneficial to know if the results would be replicated within a community clinical setting, as our study was conducted within a university treatment research clinic.

Related to design, we had three to six data points in the baseline phase, and three in the treatment and follow-up phases. More data points in each phase, with a minimum of five in the treatment phase would be preferable. We demonstrated control for maturational effects on the selected behaviour for four of the five participants. We only demonstrated control for maturational effects for Lachlan on a stimulus control behaviour. Further studies should explore options for behaviours appropriate to monitor for maturational change, and explore stimulus generalization tasks and more complex behaviours as control measures for this purpose.

In this study, two participants demonstrated generalization to more complex speech behaviours. Further investigation of the factors associated with generalization in ReST treatment is required, and more data collection points within each phase may clarify the results. The participants in this study imitated the treatment and probe items, which may lead to limited generalization to spontaneous speech. Further investigation of the spontaneous speech production following ReST treatment is warranted.

The dependent variable in this study was the percentage of words produced with correct sounds, lexical stress and smooth connection between the syllables. It would be beneficial to investigate the change within each construct, within participant, over the course of treatment in order to provide information about precisely what changes for each child, when the change occurs, and any pattern of change. The participants in this study used headphones, which would have attenuated their auditory feedback from their own speech. This may have potentially reduced the treatment effect, but given the large treatment effects demonstrated may not be of significance on this occasion. Finally, future speech treatment studies using video conferencing should include routine testing of the bandwidth at the start of each session in order to provide information about the minimum bandwidth for effective treatment.

Conclusions

This study evaluated the efficacy of ReST treatment provided by telehealth to five children with CAS. Results showed significant acquisition of the imitated targeted pseudo-words, and generalization of the treatment effect to untreated imitated pseudo-words and real words. These results suggest that video conferencing as a service delivery method for ReST treatment may be beneficial for children with CAS. These results warrant larger scale studies.

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Notes

1. For children ≥ 11 years, the stimuli of the DEAP (Dodd *et al.* 2006) were not considered sufficiently challenging to assess inconsistency. For these children an inconsistency measure was calculated for 25 words from the Polysyllabic Word Test (Gozzard *et al.* 2006). As there are no guidelines for the severity of inconsistency with these stimuli, we assumed children of 11 years would show $< 30\%$ inconsistency. For children < 11 years we used the 40% criteria for inconsistency, as reported by Dodd *et al.* (2006).
2. Due to logistical constraints, the first author, experienced in ReST treatment via telehealth, conducted one treatment session of both Jack and Emily's 12 treatment sessions.

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Supporting Information

Additional Supporting Information may be found in the online version of this article at the publisher's website:

Appendix 1 Probe stimuli for the children