INTRODUCTION

Phonological alexia is characterized by a greater deficit in sounding out pseudowords than reading real words (Beauvois & Derouesne, 1979). Poor phonological awareness is also commonly reported in phonological alexia, resulting in difficulty identifying the number, order and sameness or difference of phonemes in words on auditory tasks (Mitchum & Berndt, 1991). From the phonological deficit hypothesis, deficient phonological awareness may impair reading, spelling and speech skills (Rapcsak et al., 2008). If treatment of alexia is not initiated, then reading deficits persist (Behrmann, Black, & Bub, 1990; Wilson, 1994). However, while neurorehabilitation of phonological alexia has some positive findings, poor treatment generalization to non-treated stimuli is common (DePartz, 1986; Kendall, McNeil, & Small, 1998; Kendall et al., 2006; Kim & Beaudoin-Parsons, 2007; Kiran, Thompson, & Hashimoto, 2001; Matthews, 1991; Mitchum & Berndt, 1991; Nickels, 1992; Wilson, 1994). Also, little is known about alexia treatments' impact on functional reading skills or performance on nationally normed standardized measures of reading (Cherney, 2004). Thus, despite progress in the neurorehabilitation of alexia, significant obstacles remain for maximal recovery of functional reading skills.

Recent evidence from behavioral and neuroimaging studies (Foxe et al., 2002) have expanded earlier evidence of multisensory features of phonological processing (Ojemann & Mateer, 1979). Phoneme perception and phonological awareness both develop during speech perception and production activities, well before learning graphemes (Kuhl et al., 2007; Kuhl et al., 2006; Torgesen, Wagner, & Rashotte, 1994). Learning to perceive and produce phonemes may be derived from multisensory experiences with simultaneous or sequentially paired neural inputs from visual, auditory, oral articulatory and motor articulatory systems (Hickok & Poeppel, 2007; Kuhl et al., 2007; Pulvermuller et al., 2006). Each sensory and motor input may vary in its salience to the development of phonological processing. Likewise, the relative degree of salience for one sensory input versus another may vary from person to person (Foxe & Schroeder, 2005). Growing evidence of multisensory features of speech perception and production raises the question of whether a multisensory or multi-modal treatment program may help improve impaired phonological awareness and phonological alexia. Overall, neurodevelopmental models of language (Alexander & Slinger-Constant, 2004) and empirical studies of language development and function provide a reasonable rationale for a novel approach using multi-modal phonological perception, production, reading and spelling activities to rehabilitate phonological processing and reading skills. The present study explored the impact of a multi-modal treatment of phonological processing and reading by exploring the following questions:

1. Can multi-modal associations of phonemes be successfully trained?
2. Can phonological awareness be improved?
3. Can nonlexical reading skills be improved?
4. Does improved nonlexical reading generalize to improved lexical reading?
5. Will treatment and generalization effects be maintained three months post treatment?
6. Does Functional Magnetic Resonance Imaging (fMRI) of pseudoword reading and repetition show evidence of neural reorganization consistent with behavioral improvements?

METHODS

Participants

Participants were three males, two females, average of 53.6 years old, 66.8 months post stroke onset, 12.4 years education, with single left hemisphere strokes (documented by imaging), >6-months post-stroke, right handed, monolingual English, and aphasic (Tables 1 & 2). Exclusion criteria included significant apraxia of speech, untreated psychiatric illness, neurological illnesses, chronic medical illness, and severe impairment in vision or hearing. All participants provided informed consent via protocol approved by an Institutional Review Board, and were recruited through a VAMC.

Participants completed standardized tests of linguistic and phonologic functions at pre- and post-treatment. Participants demonstrated: (1) anomic aphasia (this alexia study was appended to a larger anomia treatment study); (2) sufficient auditory comprehension; (3) score < 45 on Boston Naming Test (Kaplan, Goodglass, Weintraub, & Segal, 1983); and (4) existing phonological functions (Comprehensive Test of Phonological Processing) (Wagner, Torgesen, & Rashotte, 1999); (Lindamood Auditory Conceptualization Test) (Lindamood, 1985).

INSERT TABLE 1 HERE

INSERT TABLE 2 HERE

Treatment procedures

Treatment was administered 2 hours/day and 4 days/week for 12 weeks (96 hours). Ttreatment trained multi-modal representations of English phonemes (associating acoustic properties, line-drawings of the oropharyngeal articulatory apparatus [mouth], proprioceptive and visual feedback from their own phoneme productions, and verbal labels of distinctive oral-motor features of each phoneme). Second, treatment used multi-modal representations of phonemes to train phonological awareness, reading and spelling skills. Hence, treatment progressed from isolated phoneme perception/production tasks to simultaneously training phoneme segmenting (spelling) and blending (reading) tasks, with one to five phoneme pseudowords (V to CCVCC) and one to three syllables. Treatment stimuli progressed from using concrete mouth line-drawings to represent phonemes to colored blocks and pieces of felt for nonlexical phonological awareness training. Later graphemes were used for continued pseudoword reading and spelling training. Throughout treatment, multi-modal features of phonemes (acoustic, verbal labels, proprioceptive feedback, visual feedback,…) were queried to aid performance on problem-solving activities of phonological awareness, reading and spelling tasks.

Treatment stimuli

Trained English graphemes included vowels (ee,i,e,ae,a,u,o,oe,oo) and consonants (p,b,f,v,t,d,k,g,th,th,s,z,sh,zh,ch,j,l,r,w,h,wh,m,n,ng), and their most common phoneme; with a limited set of vowels treatment focused on training skills with pseudowords of an increasing numbers of phonemes.

Experimental Design

A single-subject ABA repeated-probe design with replication across 5 participants, and with pre- and post-treatment standardized testing of real word reading, pseudoword reading and phonological awareness measures was employed.

Eight baseline data points were collected before treatment for the treatment probe (production of trained phonemes), generalization probe (pseudoword repetition) and control probe (Test of Nonverbal Intelligence-TONI) (Brown *et al.*, 1990). During 96 hours of treatment, repeated probes were administered every 8 hours (twelfth data point was not collected). Also, repeated probes and standardized tests were administered one week and three months post-treatment. Pilot fMRI of pseudoword reading and pseudoword repetition occurred for one participant at pre and post-treatment.

Outcome Measures

The following data was collected to answer the respective experimental questions:

1. Repeated probes of phoneme production
2. Pre and post-treatment standardized testing of phonological processing (CTOPP)
3. Pre and post-treatment standardized testing of nonlexical reading (WRMT-R; Word Attack)
4. Pre and post-treatment standardized testing of lexical reading (WRMT-R; Word Identification)
5. Probes and standardized tests were repeated at one week and three months post-treatment.
6. fMRI of pseudoword reading and pseudoword repetition

RESULTS

Judges determined all participants showed marked improvement in phoneme production (Figure 1), a few improved in pseudoword repetition (Figure 2) and none changed on control task (Figure 3). Gains in production were not maintained at follow-up, but gains in pseudoword repetition were maintained and improved for one participant. Figures 4, 5, 6, and 7 show variable improvement on standardized tests of Pseudoword Blending, Pseudoword Segmenting, Real Word Blending, and Real Word Segmenting, respectively; at least one standard deviation of improvement was considered clinically significant (see asterisks). Two participants improved on 3 phonological tests and another participant improved on two (most impaired at pre-testing); all gains were maintained at 3-months. For all participants, pseudoword reading improved with treatment stimuli; some participants achieved advanced levels of training (3-4 syllable pseudowords) and others did not (1-2 syllable pseudowords). However, no participants improved on standardized testing of pseudoword reading.

Pilot fMRI of overt pseudoword reading and repetition for one participant showed increased left-hemisphere perilesional activity in residual language cortices and reorganization in right-hemisphere language homologues (Figures 8 and 9).

INSERT FIGURES 1-9 HERE

DISCUSSION

The current pilot study explored a multi-modal treatment's impact on phonological processing and reading in phonological alexia. While all participants learned multi-modal associations of phonemes and improved isolated phoneme production skills, only three participants improved phonological processing. Participants' different levels of treatment progress may account for variable performance on outcome measures. Similarly, the disparity between improved pseudoword reading during treatment and no gains on standardized testing may be due an emphasis on multisyllable words and phonics principles that were not trained in treatment. Two participants that improved phonological processing also showed marked improvement of lexical reading skills. From the phonological deficit hypothesis and evidence of increased neural activity in residual language cortices, it is possible that the multi-modal treatment facilitated re-engagement of a lexical reading network that aided improvement on standardized tests for these two participants. Overall, these results provide preliminary evidence of treatment effectiveness and imply that behavioral improvements may be related to neural reorganization in both right and left hemisphere language regions for some individuals.

Table 1 Participant Demographic Characteristics

Subject Number	Age	Gender	Education	Duration Post Onset (months)	Lesion Localization
5	61	M	14	105	6 cm anterior-posterior diameter left MCA distribution infarct involving operculum and surrounding frontal, parietal and temporal cortex, putamen, insula and posterior two thirds of lenticulostriate endzone
6	65	F	12	16	Left putamenal hemorrhage with involvement of adjacent frontal, temporal and parietal white matter.
7	48	M	12	72	Left MCA territory infarct involving striatocapsular region, insula, and extensive portions of frontal convexity cortex
9	46	F	12	60	Left MCA aneurysmal rupture with associated 4 by 4 cm hemorrhage into putamen and deep frontal, temporal and parietal white matter
10	48	M	12	81	8 cm anterior posterior diameter left MCA infarct involving operculum and fronto-parietal convexity cortex extending up to anterior cerebral artery territory and deep to ventricular surface

Table 2 Participant Language Testing

Test	Pre-Treatment	Post-	3 month
	M (SD)	M (SD)	M (SD)
WAB Aphasia Quotient	85.2 (3.8)	90.3 (5.3)	88.2 (3.6)
Boston Naming Test	38.6 (4.6)	35.8 (11.4)	40.25 (10.3)
Controlled Oral Word Association	15.2 (10.2)	16 (11.5)	19.8 (10.6)

Figure 1 Phoneme Production Probes

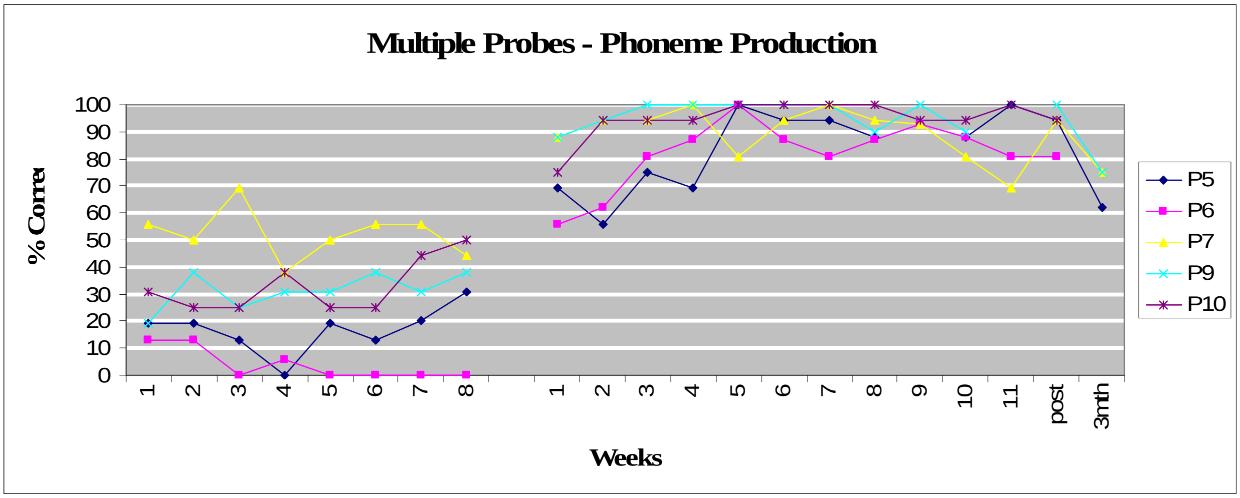


Figure 2 Pseudo Word Repetition Probes

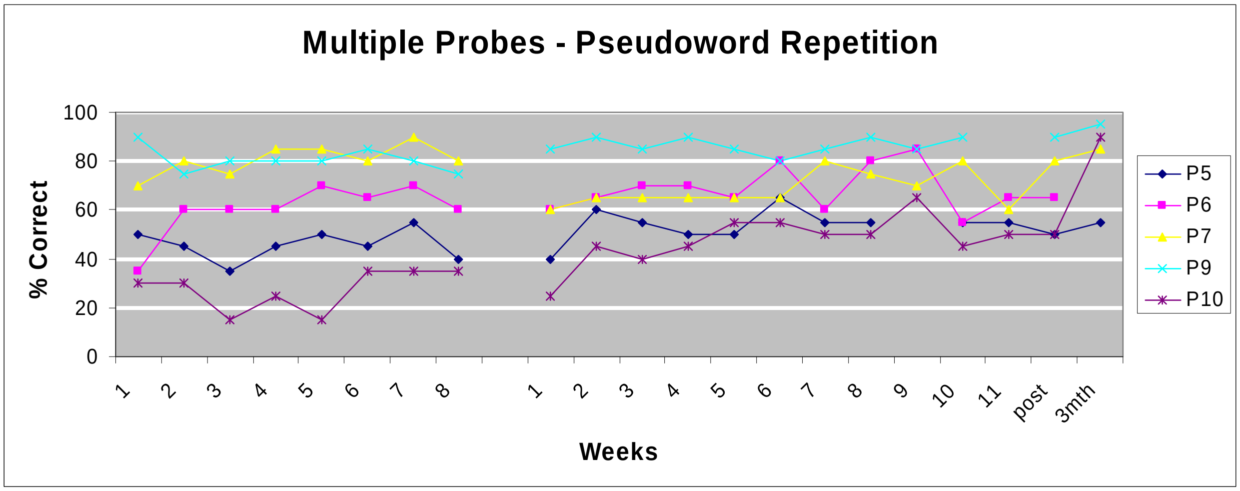


Figure 3 Control Task Probes

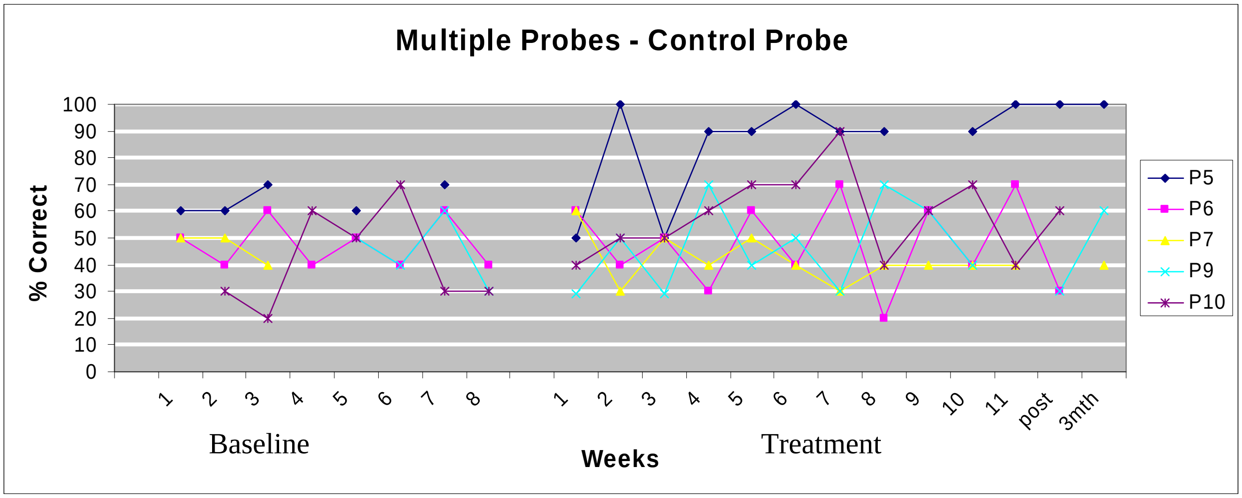


Figure 4 Pseudoword Blending – Standardized testing

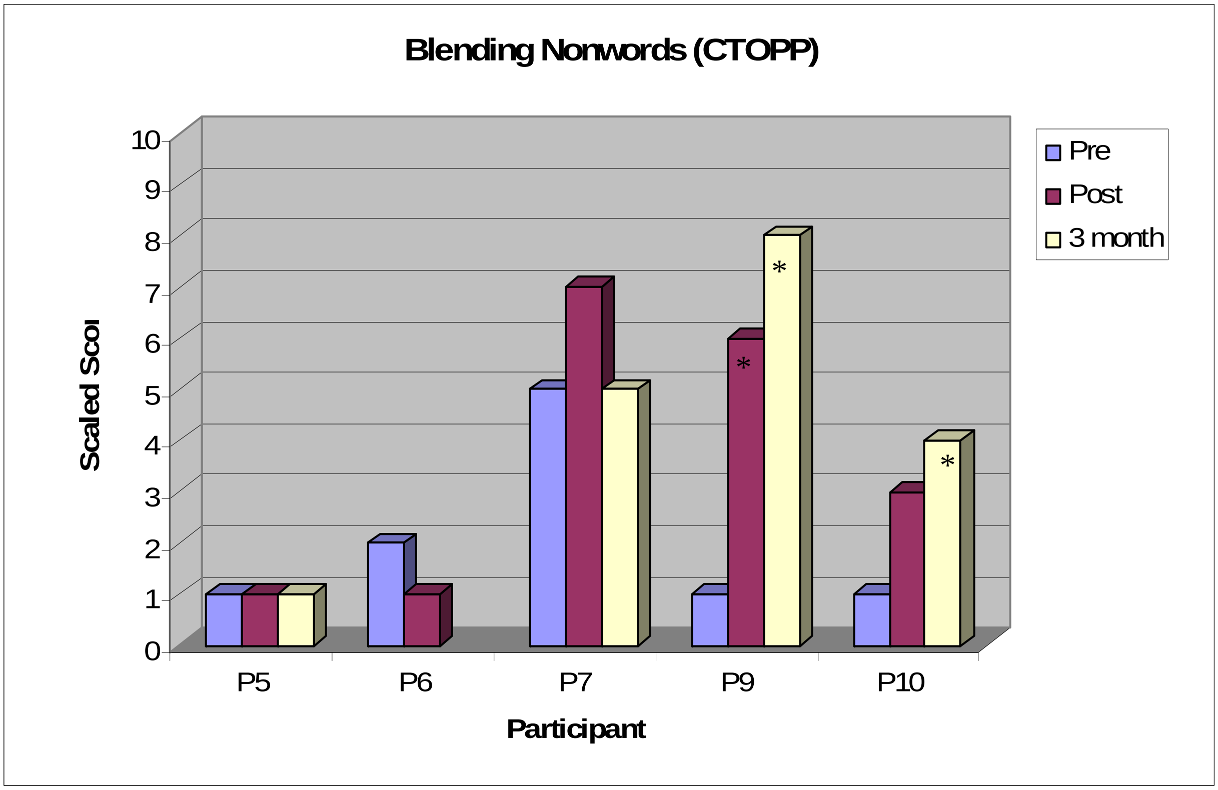


Figure 5 Pseudoword Segmenting – Standardized testing

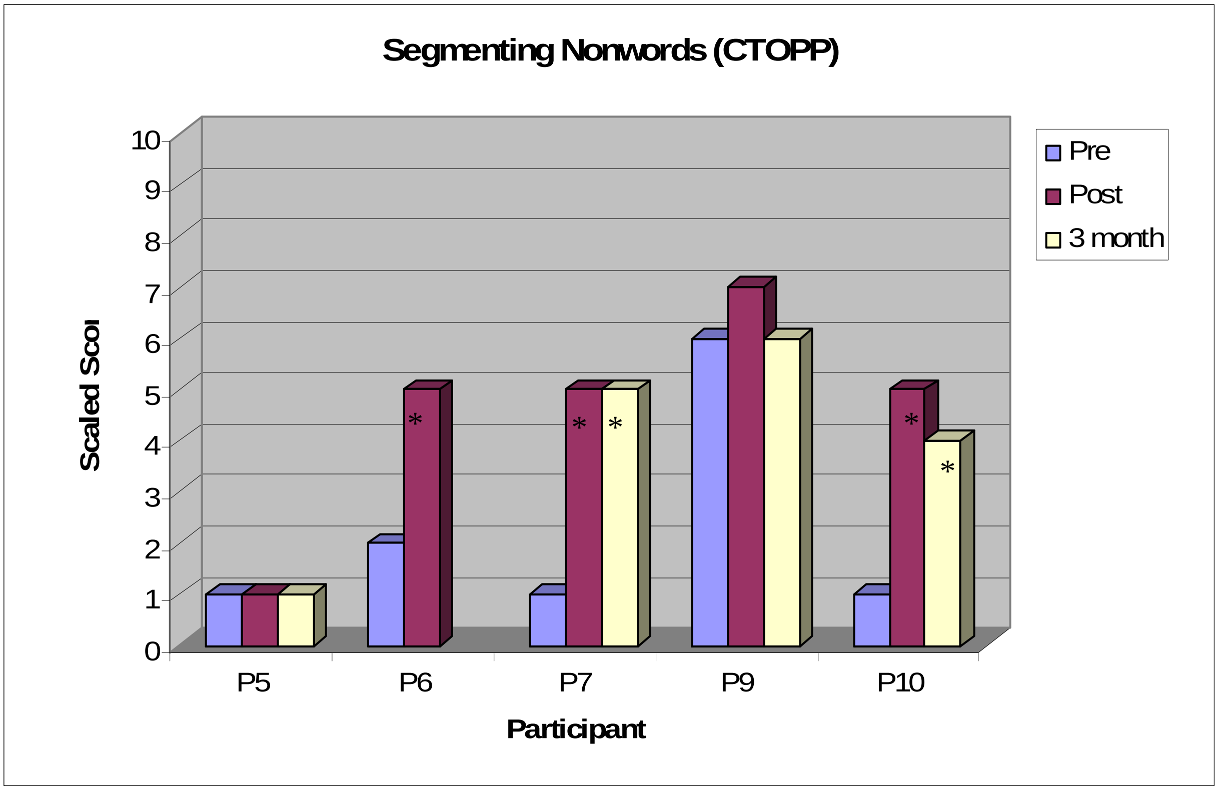


Figure 6 Real word Blending – Standardized testing

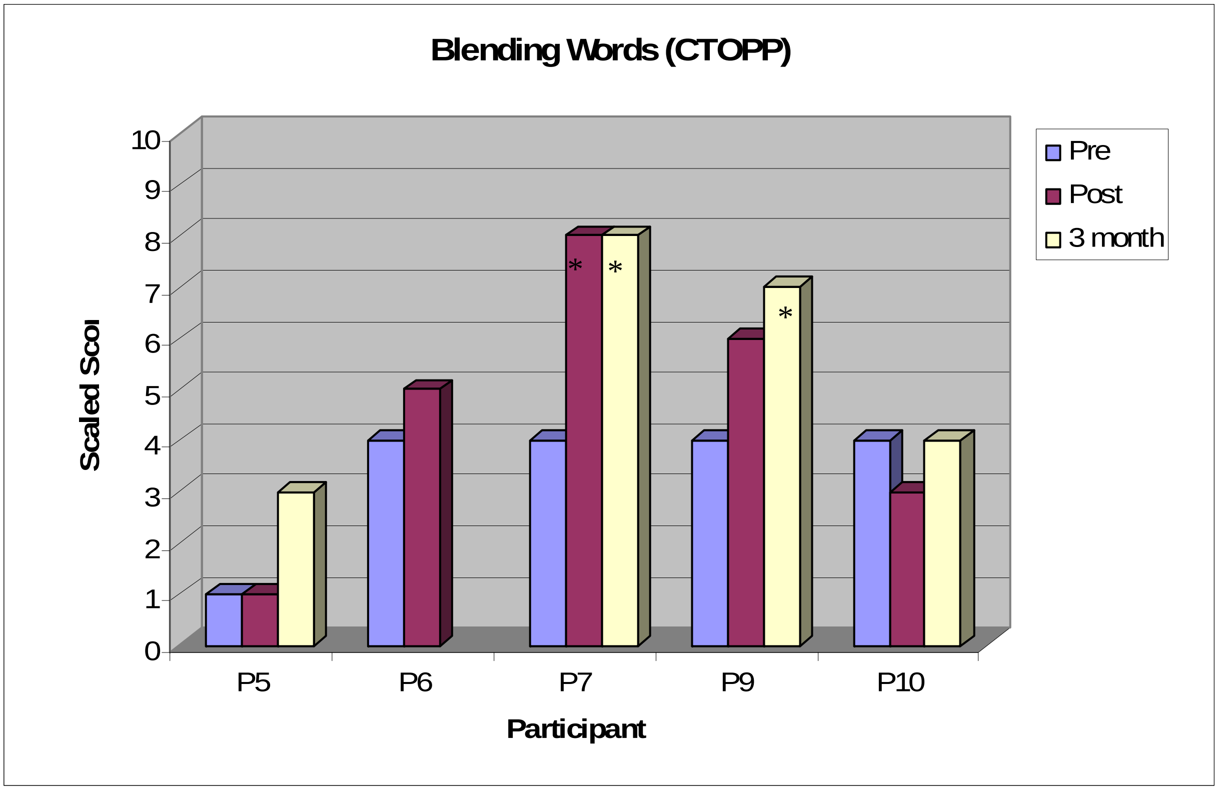


Figure 7 Real word Segmenting – Standardized testing

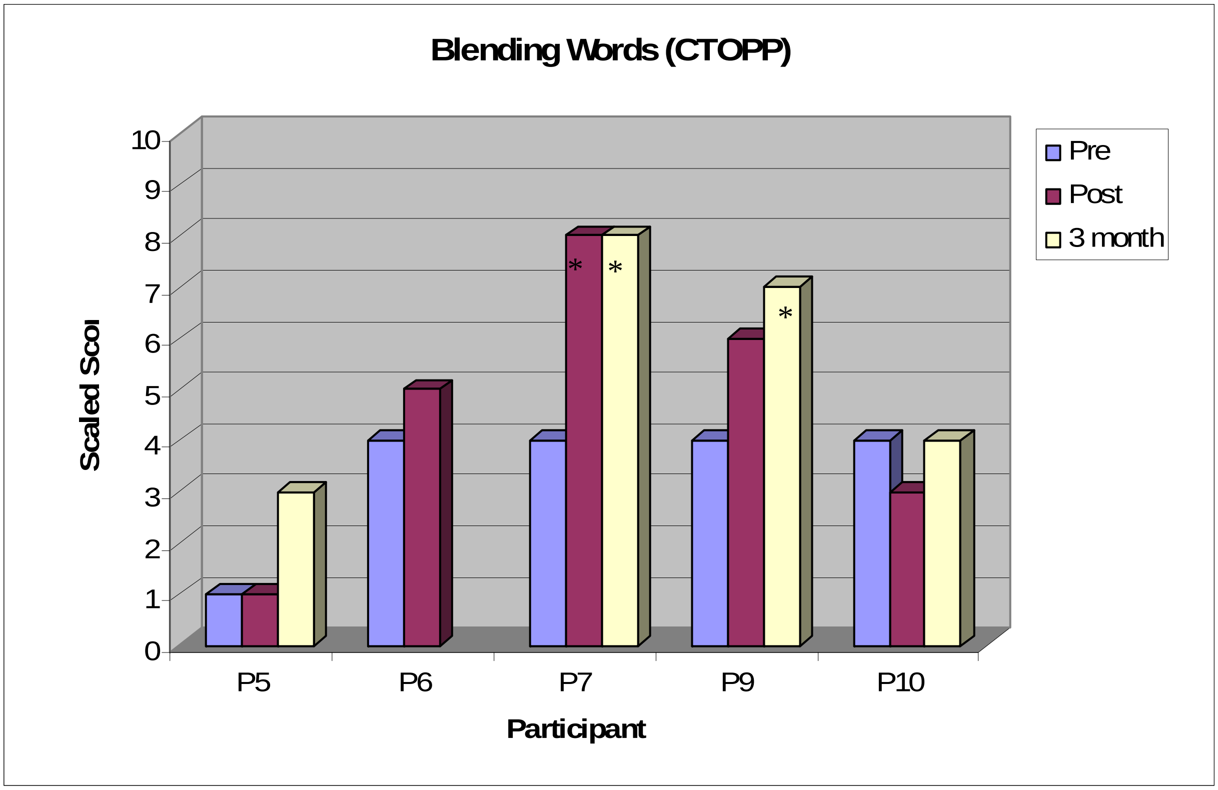


Figure 8 fMRI of Overt Pseudoword Reading for Participant #10

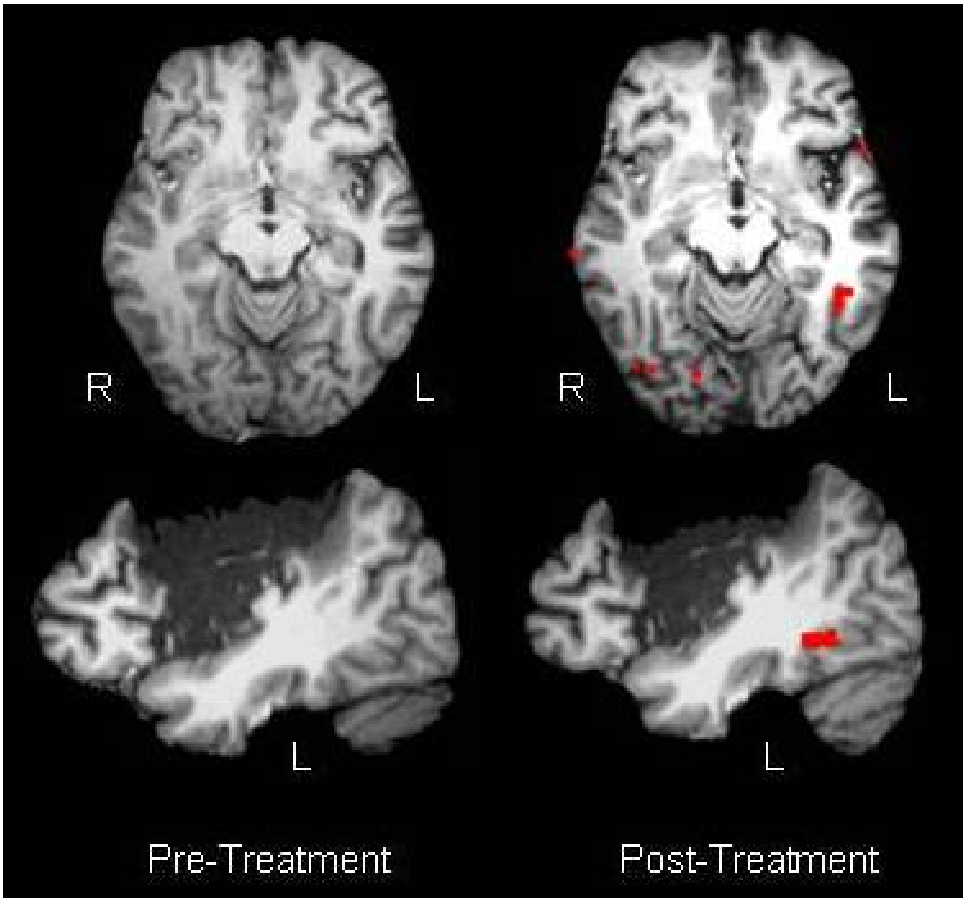
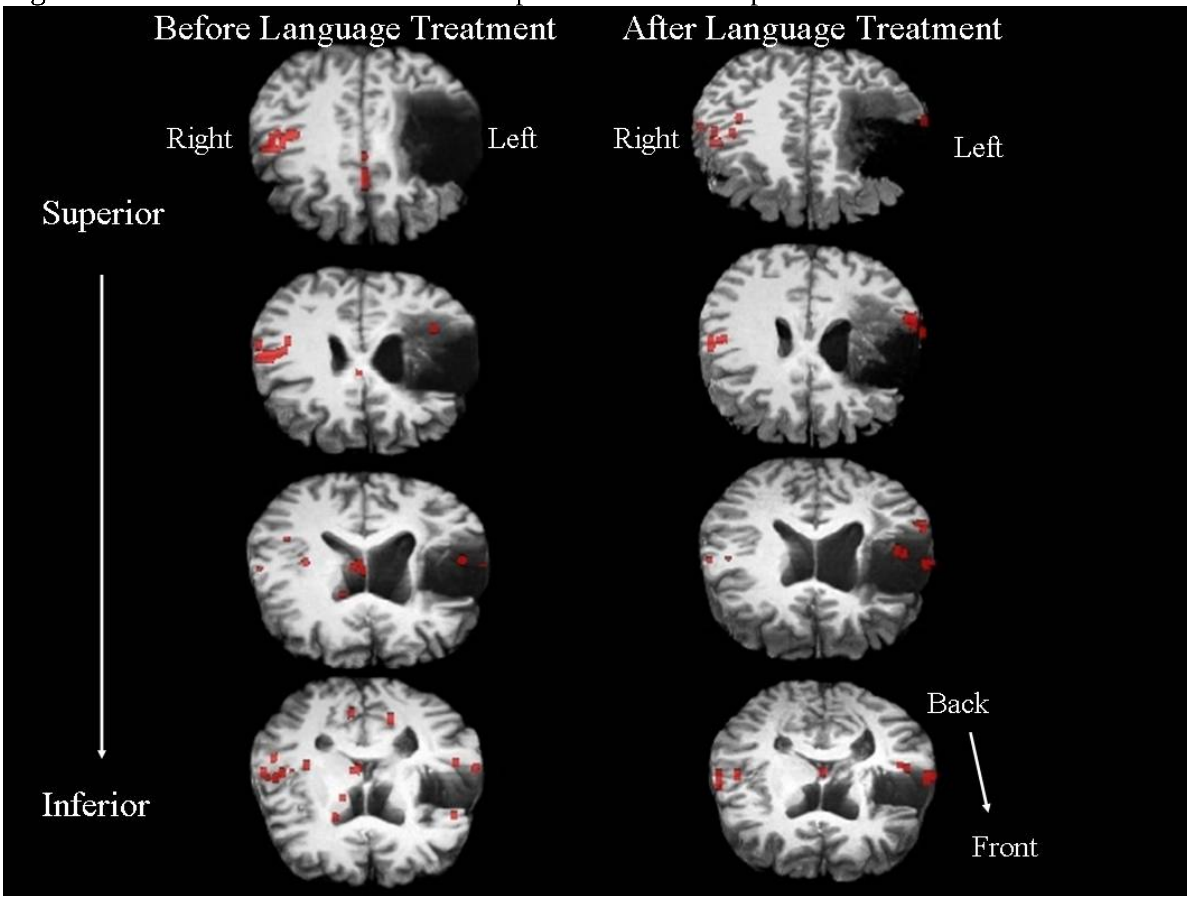


Figure 9 fMRI of Overt Pseudoword Repetition for Participant #10



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