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A Case Study of Oncological Aphasia: Clinical Profile and Response to Integrated Language Treatment

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Abstract

Purpose: A large body of research addresses the best methods and practices to treat individuals with aphasia. Much of this research focuses on individuals who have aphasia secondary to stroke. While the most common cause of aphasia is stroke, aphasia can also result from other brain diseases or injury. Relatively little research has focused on oncological aphasia resulting from brain cancer. This research examined aphasia treatment efficacy in an individual with aphasia following removal of a brain tumor.

Methods: Standardized testing was used to evaluate the clinical profile of an individual with oncological aphasia. An integrated language treatment approach was implemented with one participant with fluent aphasia using a multiple baseline across behaviors design. CIUs/utterances and percentage of CIUs produced were compared across baseline, treatment and post-treatment phases with four different conversational partners. Treatment effect size was calculated with each conversational partner. Standardized assessments were also administered before and after treatment.

Results: Small treatment effect sizes were found with three of the four conversational partners. The participant showed generalization of skills acquired on standardized motor speech, spoken language, memory, and functional communication measures.

Discussion: Findings add to evidence in support of integrated treatment approaches and add to the knowledge of the baseline performance of individuals with aphasia due to brain tumor removal. Findings suggest that effects of integrated treatment extend to functional communication.

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Introduction

Aphasia, a common neurogenic language disorder, touches many lives. An estimated one million individuals in the United States have aphasia and an estimated 80,000 new cases are diagnosed each year (American Speech-Language-Hearing Association [ASHA], 2014b). Aphasia is caused by disease or trauma to areas of the brain that are involved with language processing. Aphasia can affect any or all of the following: speaking, listening, reading, and writing (ASHA, 2014a). According to ASHA (2014b), the most common cause of aphasia is stroke. An estimated 25-40% of stroke survivors have aphasia (National Aphasia Association, 2014). Aphasia also frequently occurs as a result of dementia, traumatic brain injury, and brain tumors (ASHA, 2014a). Unfortunately, there are no available statistics specifying the percentage of people who have aphasia as a result of these conditions. However, given the high prevalence in the U.S. of dementia (5.2 million, Alzheimer’s Association, 2014), traumatic brain injury (3.2 million, Corrigan, Selassie & Orman, 2010), and brain tumor (700,000, American Brain Tumor Association, 2014), it is likely that thousands of new cases of aphasia occur each year because of these conditions.

Aphasia is a complex disorder that manifests differently depending on its etiology. The etiology of aphasia is closely tied to the demographic characteristics of patients, the nature of the neuropathology, and progression of the disorder. For example, patients who have aphasia as a result of stroke or dementia tend to be older, whereas patients who have aphasia as a result of traumatic brain injury or a brain tumor tend to be relatively young. With respect to neuropathology, stroke and tumor result in more focal lesions in the brain, whereas dementia and traumatic brain injury result in more diffuse lesions and patterns of pathology. In terms of disease progression, aphasias resulting from stroke and traumatic brain injury are acquired suddenly and will typically improve slowly over time as a result of spontaneous recovery and/or intervention. In contrast, dementia and cancer related aphasias are progressive and slow in onset. While aphasia resulting from dementia is degenerative and will typically worsen over time, aphasia resulting from brain tumor can be either rehabilitative or degenerative depending on the progression of the cancer. If the cancer is removed and does not recur, language abilities will likely improve. However, if the cancer is reoccurring or terminal the language capabilities will likely continue to deteriorate (Shafi & Carozza, 2012).

A recent study done by Davie, Hutcheson, Barringer, Weinberg, and Lewin (2009), found further differences between aphasia caused by cancer and by stroke. They found that the severity, pattern of impairment, and variability of the aphasia differed in patients who experienced stroke and those who underwent brain tumor resection. Patients evaluated after brain tumor resection had lower rates of global aphasia (3%) and higher rates of anomic aphasia (49%), while stroke patients showed higher rates of global aphasia (20–40%) and lower rates of anomic aphasia (9–28%). They concluded that “aphasia is most frequently mild and anomic after brain tumour resection regardless of lesion location, tumour grade, or awake intraoperative monitoring (p. 1205).” This contrasts with manifestations of aphasia following stroke, which is more likely to be affected by several factors, including: severity, speed of medical intervention and location in the brain.

Although aphasia symptoms and treatment prognosis vary depending on the etiology, the majority of treatment research focuses on aphasia resulting from stroke. To investigate previous research comparing treatment responsiveness in oncological vs. post-stroke aphasia an initial literature search was conducted. The search included multiple databases (MEDLINE, Academic Search Premier, ERIC, CINAHL, Google Scholar & PsycInfo) and the following search terms:

aphasia, brain tumor, brain cancer, glioblastoma, and language treatment. The majority of the published research focused on different surgery techniques and general treatment benefits without making any specific reference to language treatment. One article mentioned that participants received speech therapy if they had aphasia but did not describe the therapy or report outcomes (Bartolo et al., 2012). Only one paper was identified (Shafi & Carozza, 2012) that specifically addressed language treatment for aphasia in patients with brain tumors. According to the authors, as of 2012, there is no current research that is specific to treating aphasia in patients who have brain tumors removed. They do state in their article that patients with aphasia secondary to brain tumor removal will require treatment due to the tenacious nature of their deficits. They also suggest that terminal patients with brain cancer need the support of speech language pathologists to help them select and use an assistive device to communicate. They predicted that the number of cases of oncological aphasia seen by speech language pathologists will continue to increase as the population ages and cancer treatments improve.

To further research material available on cancer related aphasia, three meta-analyses of language treatment efficacy in aphasia were consulted (Brady, Kelly, Godwin & Enderby, 2012; Holland, Fromm, DeRuyter, & Stein, 1996; Robey, 1998). The most recent analysis only included studies with participants who had aphasia as a result of a stroke (Brady et al., 2012). The other two analyses did not specify the etiology of the aphasia (Holland et al., 1996; Robey, 1998).

Given the lack of research focused on aphasia treatment in brain tumor patients, a more generalized literature search of aphasia interventions was conducted. A MEDLINE search of 30 recent aphasia treatment studies revealed that the majority of the participants (677/683) had aphasia secondary to stroke. The remaining six participants had diagnoses of primary progressive aphasia (4 participants), traumatic brain injury (1 participant), and unspecified etiology (1 participant). Notably not a single participant in the 683 cases had aphasia as a result of brain cancer.

Given the variability across different etiologies of aphasia, unique features of aphasia secondary to brain tumor removal, and lack of available evidence based practice, we cannot definitively say if this population is inherently different and/or if they respond to aphasia treatments the same way or differently as individuals with a different etiology causing their aphasia. A study done by Davie et al. (2009) "suggest that acute post-operative language functioning may be fundamentally different in patients with brain tumours compared to patients who have had a stroke (p. 1205)." Further research is therefore needed to better understand differences in manifestations and treatment responsiveness in oncological vs. other types of aphasia. To address this need, the purpose of this research was to 1) characterize baseline performance of an individual with aphasia resulting from brain cancer, 2) evaluate the clinical efficacy of an integrated language treatment approach and 3) investigate generalization of treatment effects across a broad range of cognitive and communication measures.

Integrated treatment is an evidence-based approach to intervention that combines elements of part and whole language treatments into one treatment protocol (Milman, Vega-Mendoza, Clendenen, 2014). It is based on a part-whole learning approach that has been used successfully in other cognitive domains, such as perceptual-motor learning, procedural learning, and second language acquisition (see discussions in Milman, Vega-Mendoza et al., 2014 & Knowles, Holton, & Swanson, 2005). It is only recently that this approach has been applied to aphasia intervention. Part language treatments focus on treating a single component of language, such as vocabulary knowledge or a particular grammatical structure (Boyle, 2004). It is assumed that improvement on isolated pieces of language will spontaneously generalize to everyday language use. Whole language approaches target communication. For example, a clinician may try to increase communication of a story by encouraging participants to draw pictures to support their narrative (Lyon, 1995). In contrast, integrated language treatments target isolated language structures and then train generalization of these structures in a whole language context.

Two preliminary studies conducted using integrated treatment approaches found that five of six participants acquired the targeted structures and maintained treatment effects for at least one month following treatment (Milman, Clendenen, & Vega-Mendoza, 2014; Milman, Vega-Mendoza et al., 2014). Milman, Clendenen et al. (2014) found that "all three participants showed statistically significant improvement on more general measures of language production" such as the Western Aphasia Battery ([WAB-R], Kertesz, 2007) and the Boston Naming Test [BNT], Kaplan, Goodglass, & Weintraub, 2001). Two of the participants also generalized their production of adjectives to untreated adjectives. Milman, Vega-Mendoza et al. (2014) also found that the three participants showed significant increases in their pre- and post-treatment performance on multiple measures of connected speech. Specifically, significant changes were noted on mean length of utterance (MLU), noun–verb ratio, open–closed class ratio, and the WAB-R Aphasia Quotients (Kertesz, 2007).

In order to maximize generalization of treatment to everyday language use, we developed a patient-centered integrated treatment approach. We began by asking our client about the impact of aphasia on his life and what he would most like to work on during treatment. He stated that he wanted to improve his ability to converse with his two daughters and also to be able to talk with his friends about sports. Next we administered an extensive testing battery to assess his cognitive-communicative abilities, focusing on those skills most relevant to his stated treatment goals. Based on his goals and our comprehensive test battery we developed an integrated part-whole conversation treatment specifically designed to improve his ability to converse with his friends about sports and with his daughters about their interests.

Methods

Participant

BG, a 35 year old, right-handed, monolingual, English-speaking male with mild, fluent aphasia participated in this study. He was diagnosed with aphasia by a previous clinician using the WAB-R (Kertesz, 2007). At the time of this study he was 5 years post re-removal of a left frontal lobe glioblastoma and hence past the period of spontaneous recovery (Culton, 1969). Since then he has received speech therapy at the Utah State University Speech-Language-Hearing Clinic for 9 months (in 3 month treatment blocks). He had previously received speech therapy at Logan Regional Hospital. BG received a high school education and also attended community college pursuing a teaching certificate before his surgeries. Prior to his surgeries he was working as an electrical technician. After his surgeries he worked at a dairy farm from fall 2012 to summer 2013 but was unable to continue due to a seizure. He was not currently employed at the start of this study. BG lives with his wife and two daughters aged 8 and 10. BG also passed a pure tone hearing screening at 20dB SPL at 500, 1000, 2000 and 4000 HZ. Prior to his brain surgery, he had no previous history of language, learning, psychiatric or neurological impairment.

Standardized Assessments

A battery of standardized assessments was administered to the participant prior to beginning treatment and immediately after concluding treatment. These assessments measured his motor speech (Motor Speech Screen, Duffy, 2013; *Apraxia Battery for Adults-Second Edition*

[ABA-2], Dabul, 2000), naming ([BNT], Kapalan et al., 2001; *Psycholinguistic Assessments of Language Processing in Aphasia* [PALPA], Kay, Lesser, & Coltheart, 1992), spoken language production abilities (*Scales for Language Rehabilitation* [SLR], Milman, in development), general language capabilities (WAB-R, Kertesz, 2007), general cognitive status (*Scales of Cognitive and Communicative Ability for Neurorehabilitation* [SCCAN], Milman & Holland, 2012; *Coloured Progressive Matrices* [RPCM], Raven, Raven, & Court 1998) learning capabilities, (*California Verbal Learning Test: Second Edition* [CVLT-II], Delis, Kramer, Kaplan, & Ober, 2000), memory (Digit Span Forwards and Reverse, Lezak, Howieson, & Loring, 2004), and functional communication (*Assessment for Living with Aphasia* [ALA], Kagan et. al., 2010; *Communicative Effectiveness Index* [CETI], Lomas et al., 1989).

Treatment Stimuli

Word Level Intervention: A total of 78 words (40 related to sports; 38 related to school activities) were trained during this study (see Appendix A). Words were chosen by the participant and the two therapists conducting the treatment. All words were nouns and ranged from 1 to 5 syllables and 3-14 phonemes. Color pictures representing the nouns were found using a Google internet search and inserted into a template styled after Boyle's (2004) semantic feature analysis template.

Topic-comment Intervention: 10 topic statements or questions were generated by the therapists to elicit a response from the participant each therapy session. Topic statements or questions were related to either sports (e.g. "I hope the Seahawks win the super bowl this year.") or his daughters' interests (e.g. "I learned a new routine in tumbling today!"). These topic-comment statements were based on BG's interests. For both conversational topics the topic-comment statements or questions ranged from 3 to 14 words in length.

Conversation Level Intervention: The participant selected a conversation topic related to either sports (during sports conversation treatment) or school activities (during school conversation treatment). If the participant did not spontaneously generate a topic, he was asked if he wanted to practice one of the topic comments for more than one conversational turn or the clinician suggested a choice of topics (e.g. "Do you want to talk about the Aggie's game last night or your last fishing trip?"; "Do you want to talk about your daughter's tumbling class or their homework?").

Experimental Design

A single subject multiple baseline design across behaviors was used to examine the effectiveness of the treatment protocol. Two one-month treatment blocks were administered consecutively. The first treatment block targeted conversations about sports and the second treatment block targeted conversations about school activities. Conversational abilities of the participant were probed throughout treatment.

Probe Procedures

A total of 34 conversational probes (19 related to sports; and 15 related to his daughters' interests) were recorded using a Sony digital recorder throughout baseline, treatment, and post-treatment phases. All conversations were five minutes in length and were conducted with one of four conversation partners. Two of the conversational partners (both matched for age, education, and sports interests with BG) participated in the sports conversations throughout the study, and two of the conversational partners (matched for age, education, and interests with BG's daughters) participated in the school conversations. Conversations were recorded at least once every two weeks with each of the four conversational partners. The participant and the conversational partner were told to have a conversation about their respective topics. No other information or structure was provided. Although conversations were recorded, the clinician was not present so that the interaction was solely between the participant and conversational partner. Each of the conversations was transcribed using standardized procedures (Nicholas & Brookshire, 1993).

Pre-Treatment Probes: A total of 7 pre-treatment probes were administered. Four of the conversations focused on sports (two partners x two weeks) and three (two partners in week 1, one partner in week 2) focused on BG's daughters' interests. A fourth conversational sample on the daughter's interests was not obtained due to a scheduling conflict.

Treatment Probes: A total of 20 conversation probes were administered with the four conversation partners throughout the treatment phase (Sports =11, Daughters' Interests =9)

Post-Treatment Probes: A total of 7 conversation probes were collected after treatment (two conversations with three of the partners and one conversation with the fourth partner).

Intervention

Four 50-minute treatment sessions were administered per week for eight weeks. With the exception of the first week of sports treatment (in which there was no conversational practice), all sessions included word production (15 minutes), topic-comment training (15 minutes) and conversational practice (15 minutes). A detailed description of the three therapy tasks is provided in Appendix B.

Reliability

Coding reliability (tally of correct information units (CIUs), Nicholas & Brookshire, 1993) was assessed for eight (two verifications x four conversational partners) of the 38 conversational probes (20%). Mean point-to-point agreement between the primary coder and the second coder for the eight conversational probes was 89.99% (range = 85.7% to 92.1%).

Analyses

The primary outcome measure for this study was CIUs/utterance produced by BG in each of the conversational samples. The proportion of CIUs produced by BG and his conversational partner were also compared. Guidelines from Nicholas & Brookshire (1993) were used to identify CIUs. Effect size was calculated with each conversation partner using guidelines and interpretation outlined in Robey, Schultz, Crawford, and Sinner (1999). Normative data were also used to compare pre- and post-treatment performance on standardized tests.

Results

Initial Clinical Profile

Initial performance on standardized measures is summarized in the second column of Table 1. At the beginning of treatment BG, presented with mild to moderate apraxia of speech as shown on the Motor Speech Screen (Duffy, 2013) and ABA-2 (Dabul, 2000). His naming was mildly impaired on the BNT (Kaplan et al., 2001) and on the PALPA (Kay et al., 1992). On the SLR (Milman, in development) he demonstrated difficulty producing complex sentences. His discourse on the SLR (Milman, in development) and the WAB-R (Kertesz, 2007) picture description was characterized by frequent word-finding errors, press of speech and low volume jargon. BG was only mildly impaired on the SCCAN (Milman & Holland, 2012) and showed above average nonverbal reasoning skills on the RCPM (Raven et al., 1998). Performance on digit span was characterized by a discrepancy between digit span forward (consistently accurate for 5 digit sequence) and digit span backward (consistently accurate for 2 digit sequence) suggesting a mild-moderate impairment of verbal working memory. BG demonstrated an ability to learn on the CVLT-2 (Delis et al., 2000), albeit at a slow rate with multiple repetitions. He also demonstrated that he was susceptible to interference effects. His functional communication measures (ALA, Kagan et al., 2010; CETI, Lomas et al., 1989) showed that he was most dissatisfied with his life in the area of conversation and feeling in control of his life. His spontaneous conversation was characterized by mild press of speech and word finding behaviors (false starts, fillers, pauses, and paraphasic errors).

Treatment Results

Figures illustrating BG's performance with each conversational partner are included below in two graphic displays. The line graphs show the mean number of CIUs produced by BG per utterance (y-axis) during baseline, treatment, and post-treatment phases (x-axis). The values between the two vertical blue lines represent the treatment phase for the targeted conversational topic. The pie charts illustrate the respective conversation load carried by BG and his conversational partner (percent of CIUs produced in the conversation by each conversational partner). Ideally, we would expect the two conversational partners to share equally in the conversational load, with each participant carrying 50% of the content of the conversation. BG is always represented by the blue portion of each chart.

Sports Conversation Partner 1(see Figures 1 and 2): BG maintained a relatively stable baseline performance (3.9, 3.5 CIUs/utterance). With respect to conversational load, BG produced approximately one third of the total CIUs (39%), whereas his conversational partner produced the majority of CIUs (61%) in the sample. During the treatment phase, BG increased his production of CIUs/utterance (6.1 CIUs/utterance at T2) and the proportion of total CIUs that he contributed to the conversation (5% increase relative to baseline performance). After sports conversational treatment ended, BG's performance dropped on both measures. Comparison of pre- and post-treatment performance was consistent with a small treatment effect (d=1.6).

Figure 1. CIUs/Utterance: Sports Conversations – Partner 1.

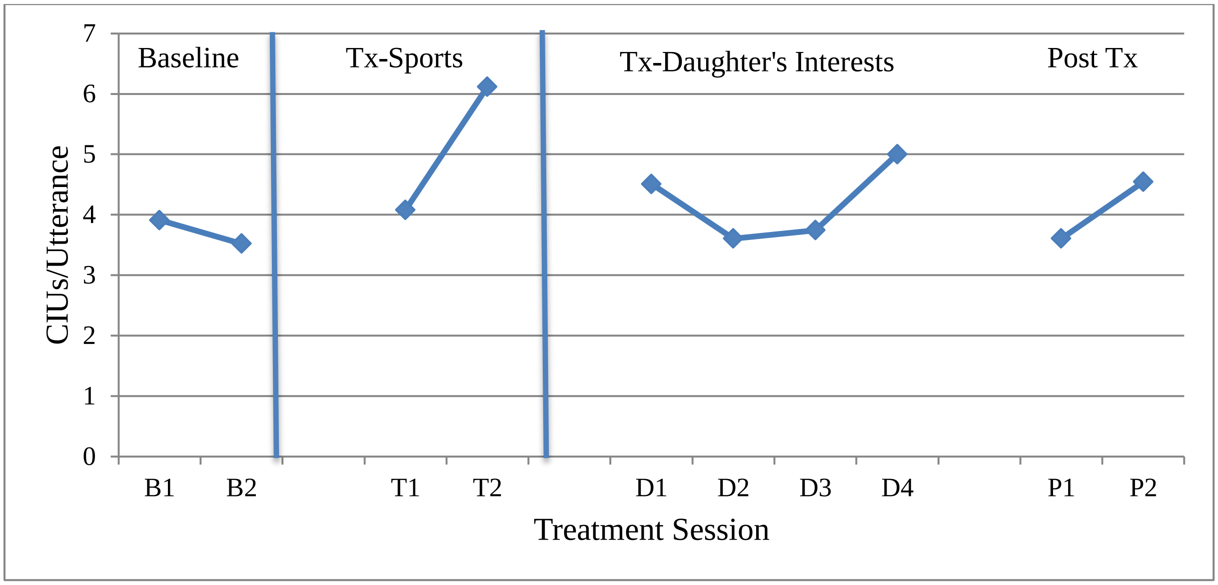
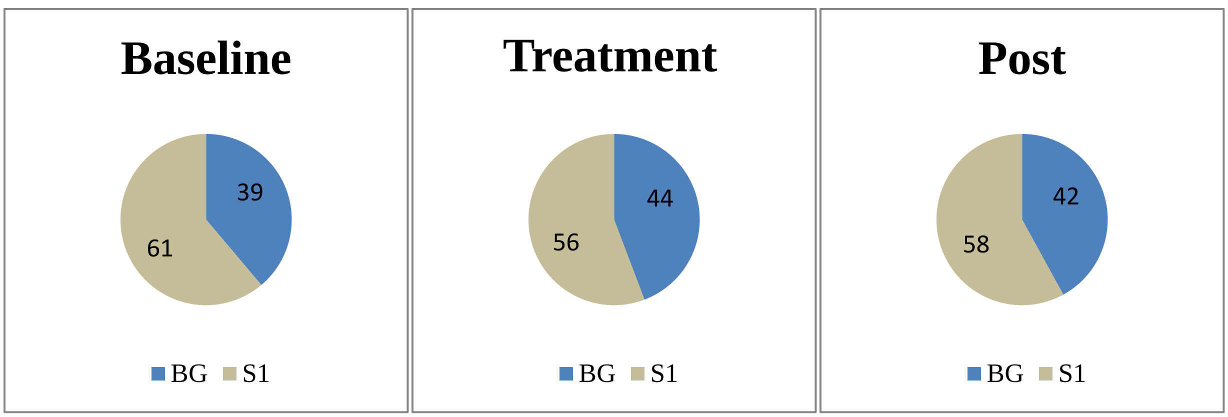


Figure 2. Percent of CIUs Produced by BG and Sports Conversational Partner – 1.



Sports Conversation Partner 2 (see Figures 3 and 4): With this conversational partner, BG's performance did increase between the two baseline measures starting out at 3.0 CIUs/utterance and then ending at 4.8 CIUs/utterance. In regards to conversational load at baseline, BG produced 42% of the CIUs in the conversation. BG's performance continued to increase in the sports phase of treatment, changing from 5.6 CIUs/utterance at week 2 to 7.1 CIUs/utterance at T2. Conversational load also increased to the point that BG was carrying more of the conversational load (64%) than his partner (36%). After treatment, BG's performance dropped on both measures to 4.3 CIUs/utterance and 49% of the conversation load. Comparing pre- and post-treatment production of CIUs/utterance revealed a small treatment effect size (d=0.8).

Figure 3. CIUs/Utterance: Sports Conversations – Partner 2.

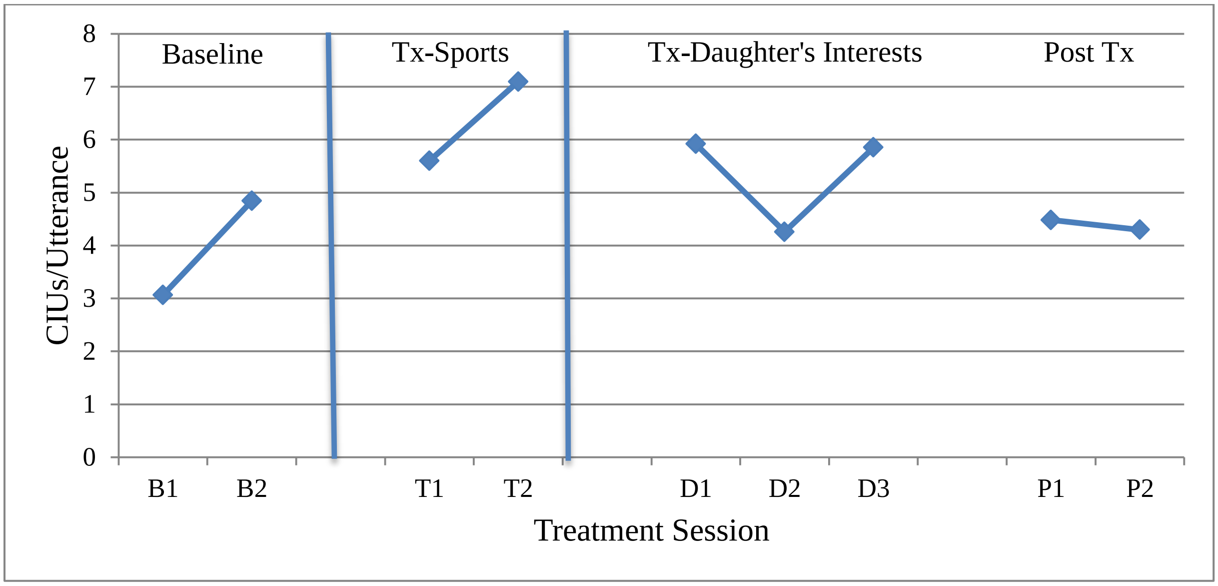
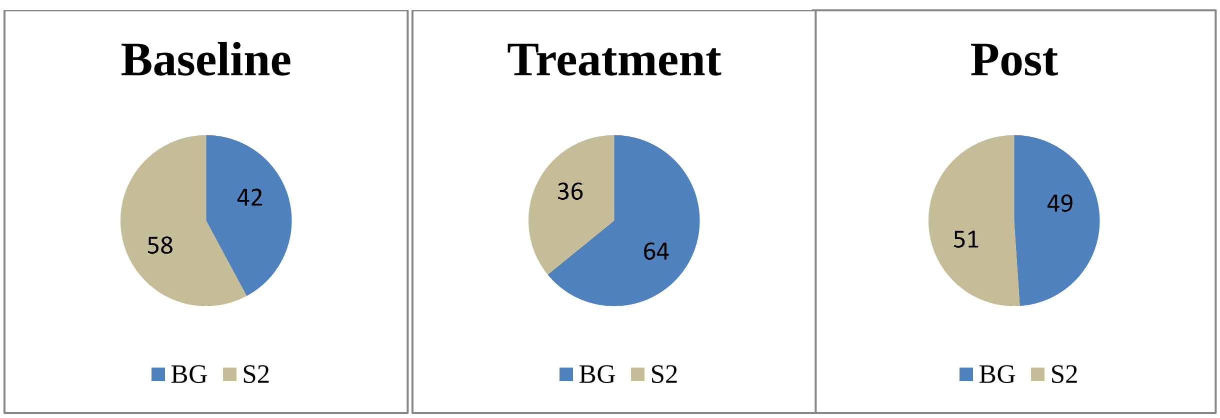


Figure 4. Percent of CIUs Produced by BG and Sports Conversational Partner – 2.



Daughter's Interests Conversational Partner 1 (see Figures 5 and 6): BG's baseline performance initially increased (2.6 -3.6 CIUs/utterance) and then leveled off by the end of the baseline phase (3.9 CIUs/utterance). With respect to conversational load, BG initially produced approximately one quarter (26%) of the CIUs compared to three quarters (74%) produced by his conversational partner. During the treatment phase for this conversational topic, BG's performance increased from 3.6 CIUs/utterance at T1 to 4.6 CIUs/utterance at T3. The conversational load carried by BG also increased to 47%. There was only one post-treatment probe with this conversational partner which remained relatively unchanged from treatment levels of performance (4.4 CIUs/utterance).   Conversational load (48% of CIUs) also remained stable relative to treatment levels of performance. Treatment effect size was small (d=1.6).

Figure 5. CIUs/Utterance: Daughter's Interests Conversations – Partner 1.

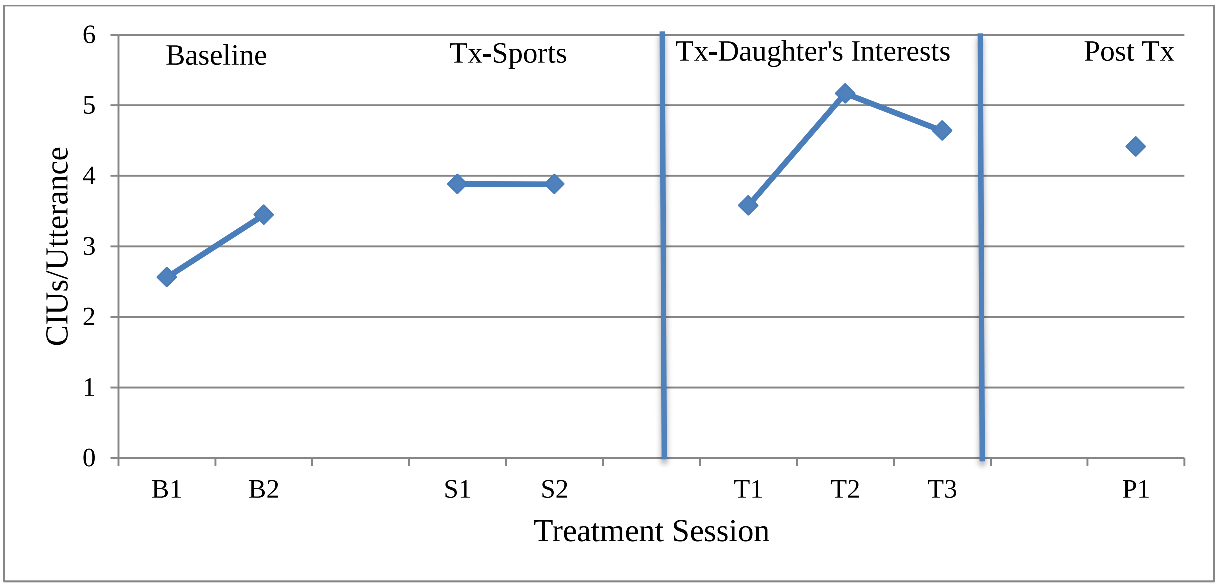
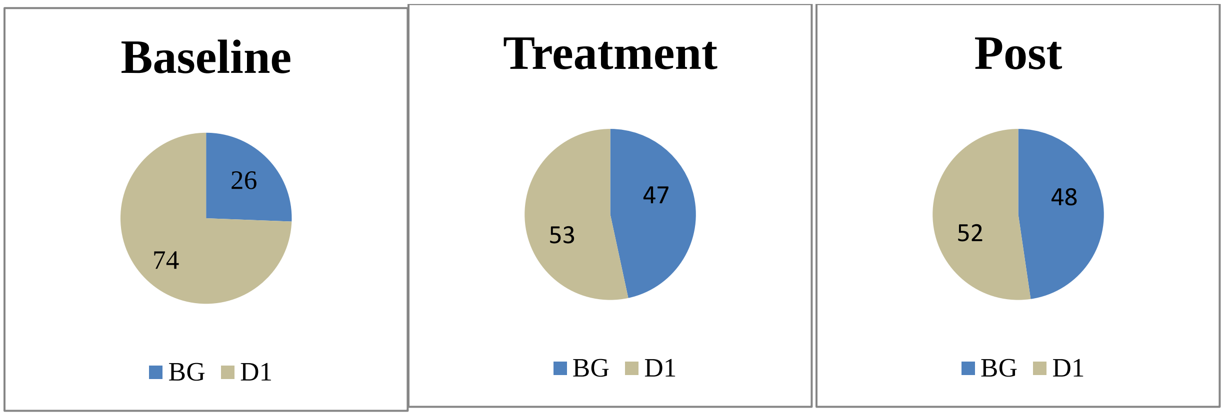


Figure 6. Percent of CIUs Produced by BG and Daughter's Interests Conversational Partner – 1.



Daughter's Interests Conversational Partner 2 (see Figures 7 and 8): At baseline BG started with 4.0 CIUs/utterance and then his CIUs/utterance spiked to 6.8 and then dropped back down to 4.1CIUs/utterance for the rest of the baseline phase. In contrast with other conversational partners, BG produced a greater proportion of CIUs (65%) than his conversational partner (35%) during the baseline phase. During treatment and post-treatment phases BG's CIUs/utterance remained relatively unchanged from his baseline performance. With respect to conversational load, CIUs initially dropped to 53% during treatment and then returned to near baseline level (69%) following treatment. There was no treatment effect size for this partner when comparing pre- and post-treatment CIUs/utterance (d= -0.7).

Figure 7. CIUs/Utterance: Daughter's Interests Conversations – Partner 2.

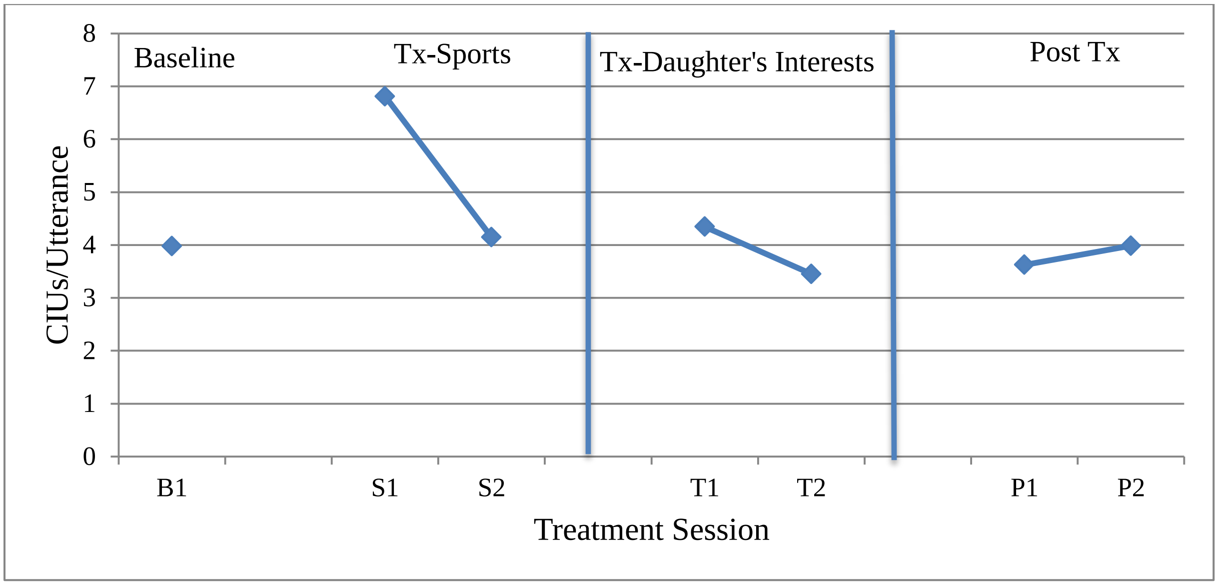
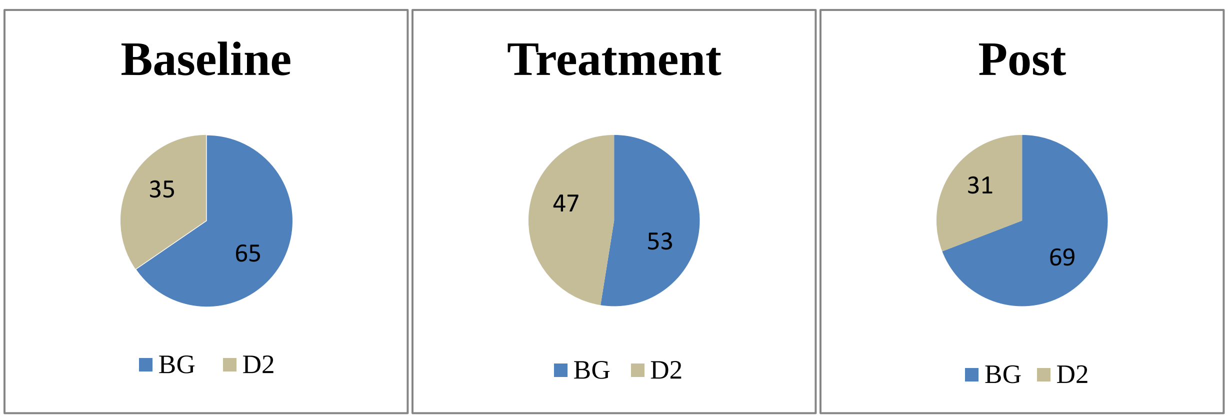


Figure 8. Percent of CIUs Produced by BG and Daughter's Interests Conversational Partner – 2.



Generalization of Treatment Effects: Results of post-testing on standardized measures are summarized in Table 1, column 3. BG performed at or near ceiling on many of the measures (BNT, PALPA, & RPCM). BG showed significant improvement on two measures of motor speech (ABA-2, Dabul, 2000): Diadochokinetic Rate (mild to no impairment) and Increasing Utterance Length (severe to mild impairment). However, his performance on the Utterance Time subtest also dropped from no impairment to mild impairment. Although normative data are currently not available for the SLR (Milman, in development), BG demonstrated qualitative gains in connected speech on this measure. Specifically, during pre-treatment performance on the sentence test BG was unable to produce any of the complex sentences. Following treatment, however, he was able to produce all complex sentences in this subtest (although half were still scored as “0” because of delayed production). CIUs produced on the discourse subtest of the SLR (Milman, in development) also increased from 55 total CIUs pre-treatment to 76 total CIUs post-treatment. Although BG’s scores remained stable on his WAB-R (Kertesz, 2007), it should be noted that the percent of utterances with word finding disfluencies (WFD) on the Picture Description task decreased from 65% with WFDs pre-treatment to 33% with WFDs post-treatment. BG’s score did not significantly change on the SCCAN (Milman & Holland, 2012) but his memory subtest score improved from 11/19 to 17/19. BG also showed improvements in verbal memory on the digit span (Lezak et al., 2004) forwards (increased from 6 to 9) and backwards (increased from 4 to 5). On the CVLT-2 (Delis et al., 2000) BG also showed an increase of approximately 1 SD on many subtests (Free Recall Correct, List B Free Recall, Total Intrusions, & Total Repetitions) and an increase of two SD on other subtests (Short-Delay Free Recall & Long-Delay Recognition False Positives). BG made significant improvements on both functional communication measures. On the CETI (pre-treatment =62.5; post-treatment = 91.25, Lomas et al., 1989) the greatest changes were seen on the questions in which he rated his conversational ability (e.g. “Being part of a conversation when it is fast and there are a number of people involved.”). On the ALA (Kagan et al., 2010), (Pre-treatment mean rating = 3.18; Post-treatment mean rating = 3.64) the most significant changes were made in participation (pre-treatment mean rating = 3.12; post-treatment mean rating = 3.74) and personal (pre-treatment mean rating = 3.36; post-treatment mean rating = 3.77) domains.

Table 1. Pre- and post-treatment performance on standardized measures.

Standardized Measure	Pre-Treatment	Post-Treatment
Motor Speech Screen (max = 69)	67	64
ABA-2		
Diadochokinetic Rate	15 (Mild Impairment)	35 (No Impairment) *
Increasing Word Length	a →8 (Severe Impairment)	a→1 (Mild Impairment)* b→2 (Mild Impairment) *
Utterance Time	11 (No Impairment)	17 (Mild Impairment) *
BNT (max=60)	51 (Mild Impairment)	53 (No Impairment)
PALPA (max=200)	193	193
SLR		
Words (max=24)	19	20
Sentences (max=20)	11	15
Discourse (total CIUs)	55	76
WAB-R		
AQ (max = 100)	89.2	89.2
SCCAN		
Total Score (max=94)	83 (Mild Impairment)	86 (Mild Impairment)
RCPM (max=37)	35	37
Digit Span Forward (max=12)	6	9*
Digit Span Reverse(max=12)	4	5*
CVLT-2 (z score)		
T1-5 Free Recall Correct	-0.4	0.3
List B Free Recall	-1.0	0.5
Short-Delay Free Recall	-1.5	1.0*
Short-Delay Cued Recall	-1.5	0.5
Long-Delay Free Recall	-0.5	0.0
Long-Delay Cued Recall	-0.5	0.0
Total Intrusions	2.0	1.0
Total Repetitions	-0.5	0.5
Long-Delay Recognition Hits	0.5	0.5
Long-Delay Recognition False Positives	2.5	-0.5*
ALA mean rating (max=4)		
Total	3.18	3.63*
Aphasia Domain	2.90	3.20
Participation Domain	3.12	3.74*
Environment Domain	3.38	3.38
Personal Domain	3.36	3.77*
Wall Question	3	3.50
CETI (max=100)	62.5	91.25*

*Denotes Statistically Significant Change

Discussion

The purpose of this study was to describe the baseline clinical profile of an individual who had aphasia secondary to a brain tumor, examine the efficacy of an integrated treatment approach, and investigate generalization of treatment effects on a broad battery of cognitive-communicative measures. In order to explore these questions a battery of standardized tests was administered pre- and post-treatment and an integrated therapy approach targeting words, single sentences, and conversation was implemented. BG made gains on trained conversational outcome measures (CIUs/utterance and conversational load) with three of the four conversational partners. In addition, his scores improved on several of the standardized measures indicating generalization of treatment effects across targeted items and contexts. Results associated with each treatment phase are discussed below.

Initial clinical profile

The first purpose of this study was to describe the baseline clinical profile of an individual who had aphasia secondary to a brain tumor versus stroke. On the WAB-R (Kertesz, 2007), BG was diagnosed as having mild anomic aphasia (AQ 89.2). This is consistent with the findings in the literature that almost 50% of individuals with brain tumor are classified as having a mild anomic aphasia (Davie et al., 2009). While consistent with brain tumors in general the fact that BG had only a mild, fluent aphasia is atypical for his type of tumor (glioblastoma). According to Whittle, Pringle & Taylor (1998; as cited in Davie et al., 2009), in patients with glioblastomas their aphasia is more severe than patients with other tumor types and severities. BG, did however, present with subtle deficits in grammar, empty speech, low volume jargon and word finding behaviors (fillers, pauses, false starts and paraphasic word errors). Although word finding errors are typical of mild anomic aphasia, the press of speech and low volume jargon were more characteristic of a severe anomia in stroke patients. Thus, while anomic aphasia can occur in individuals who have aphasia as a result of a variety of etiologies, BG's anomia was somewhat atypical of that reported for either stroke or cancer patients.

These differences in BG's presentation could be due to the slow progression of his tumor and differences in lesion location relative to aphasia caused by stroke. Davie et al. (2009) suggest "that the gradual progression of brain tumours may allow for linguistic reorganization during tumour growth that does not occur when there has been sudden destruction associated with an acute neurologic insult such as a stroke. Therefore the patient with a brain tumour may begin to compensate immediately, as soon as the first language disturbance happens (p.1204)." Other demographic and psychosocial factors, such as BG's young age and need to support a new family with young children may also contribute to his unique presentation. Since this was only a case study, it is unclear the extent to which these anomalies were due to differences in neuropathology, psychosocial variables, or the interaction of these two factors. Clearly much more research is needed to fully address these question.

Response to Treatment

Baseline: Mean baseline performance across conversational partners was approximately 4 CIUs/utterance (SD = 1.04). However there was also some notable variability in baseline measures. Three of the four participants showed an increase in the second (compared to initial) baseline measure (3 → 4.8; 2.6 →3.4; 4 → 6.8). This may have been due to a task learning effect as has been seen in previous aphasia treatment studies with high level participants (Milman, Clendenen et al., 2014; Milman, Vega-Mendoza et al., 2014). Notably, this increase was greatest for the second "Daughters' Interests" conversational partner. This may have been partially due to the fact that the second baseline measure overlapped with initiation of the sports conversation treatment. However, given the fact that all other probes with this conversational partner were close to baseline levels, it seems likely that this erratic data point was due to factors related to this particular conversational partner rather than specific treatment effects. Given the variability in baseline data observed in this and previous research, ideally baseline testing should be extended for more than two sessions. In this particular case, we were administering treatment in an active training clinic with time constraints placed on the duration of baseline performance testing. Nonetheless, the multiple baseline design allowed us to initiate treatment for Sports Conversation treatment and extend baseline testing for our second treatment replication (Daughters Interests).

Acquisition: BG showed an increase in production of CIUs/utterance and proportion of CIUs produced in conversations with three of the four partners. The fourth conversational partner remained relatively stable except for one spike in baseline. This particular partner was very young and varied in her motivation and interest in conversing with BG. While CIUs increased with the other conversational partners, performance patterns still varied across these partners. These results suggest that choice of conversational partner is an important consideration in treatment outcomes. Nonetheless, for the three partners with whom he showed an improvement in CIUs/utterance, only a small treatment effect size was observed. This again could be due to several factors: length of treatment, measuring progress with authentic and variable conversation partners, and complex treatment targets. Length of treatment could be the most significant factor for this client considering his performance on the CVLT-2 (Delis et al., 2000). He demonstrated an ability to learn but it was at a slow pace and he was susceptible to interference effects. Our treatment could have been too short of a time for BG to fully acquire the skills we were treating. We also treated two different conversation topics. It seems likely that he would have shown more improvement on a particular topic had we focused exclusively on a single topic throughout the intervention period. Manipulating these factors and assessing their effect on treatment efficacy would be an interesting topic for further research. Further research could also measure treatment effect sizes in linguistically simpler tasks targeting isolated words or sentence-level topic-comments.

Maintenance: BG's performance consistently dropped with three of the four partners once we stopped targeting the relevant treatment topic (though it remained above baseline levels). This pattern of performance is similar to that summarized in Boyle (2011) who found that not all of their participants made gains as a result of therapy and that some participants would improve right after treatment but then would return to baseline at later probe measures (p.1323). Since, we were training two different topics, the drop since seen in performance on the sports conversations could have been due to a treatment interference effect once we began training on the second conversational topic. As stated above, the treatment was administered for a relatively short duration (only one month per topic). This may not have been enough time for BG to adequately establish mastery to the level where he was able to maintain therapy levels of performance once treatment was stopped. Research has shown that therapy administered for longer duration results in larger treatment effects (Holland, Fromm, DeRuyter, & Stein, 1996). What constitutes adequate duration of therapy is less clear. Robey (1998) commented that determining the total amount of therapy needed for a client is difficult because "the severity of aphasia, the health of the patient, and the motivation of the patient" are all factors in determining the total amount of therapy needed (p. 179).

Generalization to Standardized Tests

Perhaps BG's most interesting improvement was on the standardized assessment battery that was administered pre- and post-treatment. BG improved on standardized measures of motor speech (ABA-2, Dabul, 2000), spoken language (SLR, Milman, in development), memory (digit span, Lezak et al., 2004), learning (CVLT-2, Delis et al., 2000) and functional communication (ALA, Kagan et. al., 2010; CETI, Lomas et al., 1989). All other measures remained similar to baseline performance.

With respect to apraxia, BG improved in his production of phonetically complex multi-syllabic sequences (diadochokinetic rate changed from mild to no impairment) and in his ability to produce words of increasing length (from severe to mild impairment). While he also increased his utterance time (no impairment to mild impairment), this could have been due to the fact that one of his strategies in therapy was to "stop, breathe and think." He was encouraged to speak slower to give himself the time he needed to be clearer in his speech. Gains in connected speech included improved production of complex sentences and increased production of CIUs during discourse. Although BG remained relatively stable on general cognitive measures (SCCAN, Milman & Holland, 2012; RCPM, Raven et al., 1998), his performance on measures of memory and learning (Digit Spans, Lezak et al., 2004; CVLT-2 Delis et al., 2000) improved. Improving memory and learning was not a target for this therapy and was an unforeseen outcome. One possible reason for this result could be that as his language skills improved and he gained confidence, the mental load decreased for language processing, giving BG more mental energy to focus on memory. Further research exploring the relation between integrated language treatment and verbal memory would be an exciting future direction for research.

BG also made significant improvements on both functional communication measures. On the CETI (Lomas et al., 1989) the greatest increases were seen on items related to his conversational ability. BG's most significant increases on the ALA (Kagan et al., 2010) were in the participation and personal domain. Both of these areas address how the participant feels about their life and their ability to interact with others.

In summary, results of pre- and post-testing suggest that our treatment was effective in helping BG improve his motor speech control, production of connected speech, verbal memory, and functional communication.

Comparison with Previous Integrated Treatment Approaches

It is difficult to fully compare our study with previous treatments because the primary outcome measures in earlier studies were single words and sentences (Milman, Clendenen et. al, 2014; Milman, Vega-Mendoza et al., 2014). In contrast, the primary outcome measure in this study was CIUs/utterance in a conversational context. While previous studies found significant treatment effect sizes for their outcome measures (words and sentences), the treatment effects for our study (CIUs/utterance in conversation) were only small. This difference in treatment effect size could have been due to differences in the complexity of the tasks used as outcome measures. Notably, Milman, Clendenen et al. (2014) cite the task complexity effect literature (Hartsuiker & Kolk, 1998; Shankweiler et al., 2010) to explain why their participants performed better on the linguistically simpler tasks than on more complex discourse tasks.

Although a greater number of standardized tests were used for pre- and post-testing in this study, the WAB-R, SCCAN, BNT, & CETI were also assessed in earlier work and therefore can serve as a point of comparison. In previous studies, significant increases were seen on the WAB-R (Kertesz, 2007) whereas our participant remained relatively stable on this measure (although his word-finding behaviors on the WAB-R discourse task improved). BG's performance was similar to previous research participants in showing an increase (albeit nonsignificant) on the BNT (Kaplan et al., 2001) and the SCCAN (Milman & Holland, 2012). In addition, BG showed significant improvement on the CETI (Lomas et al., 1989), while the majority of participants in previous research did not show significant changes on this measures. More modest gains on the WAB-R, BNT and SCCAN may have been due to BG's high (near ceiling) performance on these measures relative to participants in earlier studies. For instance, in earlier studies the highest aphasia quotient at baseline was 78.5 whereas BG's was 89.2. Again, differences in generalization of a treatment effect to these more general measures could also have been due to the limited time we targeted our structures, having multiple treatment topics, and the fact treatment focused on conversation which is more complex than the treatment targets in previous studies.

Clinical Implications

Though only small treatment effect sizes were achieved during this treatment, significant gains were achieved on several standardized measures which indicate overall language improvement which is consistent with previous integrated treatment studies (Milman, Clendenen et al., 2014; Milman, Vega-Mendoza et al., 2014). Importantly, significant gains were shown on both functional communication measures, especially questions measuring BG's conversational ability and his comfort level in conversations. Anecdotally, several individuals who knew BG pre- and post- treatment commented on his improved conversational skills and confidence during and after this treatment study. All four clinicians involved in this project also felt that BG made significant gains. It should also be noted that only a few months after this treatment study was concluded, BG removed himself from therapy to return to work at a job he had before his brain cancer. He himself stated several times that he felt that this therapy was the most helpful therapy he had received in terms of improving his language abilities.

Limitations and Future Directions

The results of this study are intriguing and warrant future research. This study was preliminary in nature as there was only one participant. This participant differed in important ways from individuals with aphasia secondary to stoke in his symptoms, psychosocial needs and his response to integrated treatment, especially in the area of functional communication. Replicating this study with more individuals with oncological aphasia would help to further examine individual variability in clinical profiles and responsiveness to treatment. Future research should also explore this treatment model addressing only one topic for longer periods of time to assess if duration of therapy is a defining factor.

In summary, individuals with brain tumors have a high mortality rate, but those who survive will likely be younger and will live with their aphasia for a much longer time than those who had a stroke. Learning more about specific manifestations of aphasia and responsiveness to treatment in this population will improve clinicians' knowledge and understanding of how to best serve all of their clients with aphasia regardless of etiology.

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Appendix A

Word Level Stimuli

Appendix A. Word stimuli used during treatment.

Sports	Sports	Daughter's Interests	Daughter's Interests
jig	Weston Reservoir	satyr	third base
spoons	wide receiver	front tuck	Hungry Hungry Hippos
field goal	runningback	cartwheel	Rings
New York Yankees	waders	bunt	Chores
Kapernick	flagrant foul	Elsa	tumbling gym
free throw	crossover	infielder	Librarian
incomplete pass	Jim Harbaugh	Sponge Bob	salt lick block
shot clock	Endzone	fair ball	Fan Boy and Chum Chum
hot & tots	free agent	Miss Lou	Breadwinners
blitz	three point line	Grandpa	Anna
game warden	Cutthroat Trout	Duty	Somersault
turnover	technical foul	recess	macaroni and cheese
third down	Lures	Taylor Swift	balance beam
spinning rod	Kevin Durrant	tumbling	Merida
Keeton	Assist	leotard	back handspring
nymph	Yellow Perch	sludge	round off
travelling	Nick Vigil	Kendra Sorenson	Mrs. Barton
punt	jump shot	Seth Sorenson	Granny
angler	Key		Salute
fishing Pole	alley-oop		Tim McGraw

Appendix B

Treatment Protocol

Word Level Intervention

A total of 78 words (40 sports, 38 daughter's interests) were generated. Half of the words were targeted for two weeks and then the other half was targeted. They were generated by the client and the clinician researching the treatment topic. Treatment was styled after Boyle's (2004) semantic feature analysis with consideration of Hashimoto's (2010) phonological feature analysis as phonological cues were a helpful strategy for the client. For each target word a color google image representing the noun was presented in a template styled after Boyle's (2004) semantic feature analysis template. Our client was then asked to name the word clearly and then generate the semantic category ("What kind of thing is it?"), an association ("Tell me something about it") and the first sound. Once he had generated the three features he was asked to state the target word again. If the client was disfluent during any of these steps, he was given the following cue: "*Stop, breathe, and think about what you want to say. Then say it again, nice and slowly."* Once a fluent response was produced the client was asked to write the word or the feature. If the client was unable to produce a correct feature (category, association, or sound) a multiple choice option was provided (e.g. *Is it a piece of equipment or clothing?*). If the client was unable to initially name the item, however, it was not provided until completing all of the feature prompts.

Topic-Comment Intervention

The topic-comment intervention was a modified version of Response Elaboration Training (Kearns, 1985). After research on the treatment topic the clinicians would generate a sentence or a question (i.e. "*Did you watch the opening ceremonies of the Olympics?"; "I* *learned a new routine at tumbling today."*) and the client was asked to generate an appropriate comment *("yes, I saw it"*). The clinician reinforced the client response by repeating and expanding on the original response, and by asking a follow up question *("Oh, so you watched the opening ceremonies last night, what was your favorite part?)*. After the client answered the question ("*I liked seeing the athletes"*), the clinician again repeated and expanded on the client's response, and combined it with the original utterance *("So you watched the opening ceremonies last night and your favorite part was seeing all the athletes – is that right?")*. Once the client agreed with the clinicians recast of his utterance, he was asked to repeat the expanded utterance fluently. If the response was dysfluent at any point, the client was given the cue: "*Stop, breathe, and think about what you want to say. Then say it again, nice and slowly."*

Conversation Intervention

BG would determine the conversational topic and he and the clinician would engage in more natural discourse. If he did not have a specific topic of choice he was asked if he wanted to practice one of the topic comments for more than one conversational turn or the clinician suggested a choice of topics (e.g. "Do you want to talk about the Aggie's game last night or your last fishing trip?"; "Do you want to talk about your daughter's tumbling class or their homework?"). Prior to initiating the conversation, the clinician said "*While we're talking, I want you to remember to make sure you stop, breathe, & think before you start talking. If you say something that's unclear, I'll just tell you that I didn't understand and ask you to repeat it."* During the conversation the clinicians used two levels of cueing. The first was just a simple request for clarification (*"I am sorry I didn't understand that. Can you say it again?")*. The second was using the Response Elaboration Training (Kearns, 1985) techniques of recasting to help BG expand his response (see description above). The second was used if the request for clarification was unsuccessful.