

## Longitudinal treatment of primary progressive aphasia: a case study

LAURA L. MURRAY

Department of Speech and Hearing Sciences, Indiana University,  
Bloomington, Indiana, USA

### Abstract

The purpose of this longitudinal study was to describe the evolving treatment regimen provided to a woman with a 4 year history of non-fluent primary progressive aphasia (PPA). Over her 2.5 years of treatment, three different therapy approaches were used to adapt to her changing communicative abilities and needs: (i) a traditional stimulation-facilitation approach, (ii) the 'Back-to-the-drawing board' programme, and (iii) a functional communication approach including provision of an augmentative communication device. Improvement was found following each treatment approach indicating that PPA patients can continue to benefit from long-term speech-language pathology services.

### Introduction

Since the early eighties, there has been widespread attention in primary progressive aphasia (PPA), a clinical syndrome in which patients suffer progressive language deterioration despite relative preservation of cognition, independence in activities of daily living, and unidentifiable stroke, tumour, infection, or metabolic disease (Mesulam and Weintraub 1992, Weintraub *et al.* 1990). Increased interest has produced a proliferation of case reports, most of which have addressed the continuing controversy over PPA as a separate clinical entity: whereas some researchers view PPA as simply a precursor or variant of more generalized dementias such as Alzheimer's or Pick's disease (Green *et al.* 1990, Kertesz *et al.* 1994, Poeck and Luzzatti 1988), others suggest that PPA differs sufficiently on a clinical-behavioural basis and, in some cases, on a histopathological basis as well (Duffy and Petersen 1992, Kirshner *et al.* 1987, Mesulam 1982, Weintraub *et al.* 1990). Other papers have concentrated on delineating the nature of the language impairment in PPA, which may present as a progressive fluent, non-fluent, or mixed aphasia (Delgado *et al.* 1993, Hodges and Patterson 1996, Karbe *et al.* 1993, Thompson *et al.* 1997, Tyler *et al.* 1997).

Unfortunately, treatment has been the focus of few studies. Some reports described speech-language treatment, but only secondarily to enumeration of the patient's linguistic, cognitive, and physical decline (Hart *et al.* 1997, Kesler *et al.* 1995, Northen *et al.* 1990, Scholten *et al.* 1995). Two studies utilized single-subject designs to examine the effects of treatment for PPA (McNeil *et al.* 1995, Schneider *et al.* 1996). Both studies provided detailed description of procedures and reported

Address correspondence to: Laura Murray Ph.D., Department of Speech and Hearing Sciences, Indiana University, 200 S. Jordan Avenue, Bloomington, IN 47405, USA.

Table 1. Longitudinal speech and language percentage (and raw score) data for subject D.D.

Measure	Test score at time of measurement (%)						
	April 1994	Dec 1994	Jan 1995	Sept 1995	Dec 1995	May 1996	Dec 1996
<i>MTTDA</i>							
Understanding sentences (15)	100 (15)	87 (13)					
Following directions (10)	100 (10)	90 (9)					
Repeating sentences (6)	17 (1)	17 (1)					
Reading complete sentences (12)	92 (11)	100 (12)					
Reading complete paragraphs (8)	50 (4)	50 (4)					
Oral reading, sentences (30)	93 (28)	90 (27)			27 (8)		
Repeating phrases (20)	60 (12)	45 (9)			30 (6)		
Expressing ideas (6)	100 (6)	83 (5)					
Producing sentences (6)	33 (2)	17 (1)					
Describing a picture (6)	50 (3)	67 (4)					
Naming pictures (20)	95 (19)	90 (18)					
Defining words (10)	60 (6)	50 (5)					
Producing written sentences (6)	50 (3)	17 (1)					
Writing sentences to dictation (7)	43 (3)	0 (0)					
<i>BDEA</i>							
Following commands (15)			100 (15)				
Complex ideational material (12)			75 (9)				
Confrontation naming (105)			72 (76)				
Animal naming (fluency) (19)			53 (10)				
Responsive naming (30)			100 (30)				
Oral reading, sentences (10)			70 (7)				
Repetition, words (10)			30 (3)				
Repetition, high probability (8)			0 (0)				
Repetition, low probability (8)			13 (1)				
Reading sentences, paragraphs (10)			50 (5)				
Written confrontation naming (10)			100 (10)				
Writing sentences to dictation (12)			42 (5)				
Narrative writing (4)			50 (2)				

ADP

Personal information (24)	75 (18)	75 (18)	79 (19)	75 (18)
Information units (raw score)	10	11	8	2
Naming (36)	50 (18)	69 (25)	53 (19)	33 (12)
ADP phrase length (raw score)	4.6	4.3	2	1
Auditory comprehension (28)	79 (22)	93 (26)	89 (25)	79 (22)
Repetition (36)	36 (13)	36 (13)	31 (11)	3 (1)
Writing (30)	90 (27)	100 (30)	100 (30)	100 (30)
Reading (30)	90 (27)	90 (27)	90 (27)	90 (27)
Elicited gesture (21)	100 (21)	81 (17)	90 (19)	86 (18)
Singing (9)	11 (1)	11 (1)	44 (4)	33 (3)
Lexical retrieval (standard score <sup>a</sup> )	10	10	9	8
Aphasia severity (standard score <sup>b</sup> )	100	106	103	93
Alternative communication (standard score <sup>b</sup> )	107	105	107	107
Behavioural profile (standard score <sup>b</sup> )	105	105	105	105
Aphasia type	Broca's	Broca's	Broca's	Broca's
WAB				
Aphasia quotient	67.2	69.5	60.9	50.4
Information content (10)	90 (9)	90 (9)	90 (9)	80 (8)
Fluency (10)	40 (4)	40 (4)	40 (4)	20 (2)
Complete yes/no questions (60)	95 (57)	95 (57)	95 (57)	95 (57)
Auditory word recognition (60)	95 (57)	98 (59)	98 (59)	85 (51)
Sequential commands (80)	80 (64)	80 (64)	65 (52)	65 (52)
Repetition (100)	39 (39)	45 (45)	33 (33)	23 (23)
Object naming (60)	90 (54)	90 (54)	67 (40)	60 (36)
Word fluency (20)	45 (9)	55 (11)	30 (6)	20 (4)
Sentence completion (10)	60 (6)	60 (6)	40 (4)	40 (4)
Responsive speech (10)	90 (9)	100 (10)	80 (8)	50 (5)
Reading (100)	96 (96)	84 (84)	81 (81)	84 (84)
Writing (100)	68.5 (68.5)	75 (75)	83.5 (83.5)	70.5 (70.5)
Praxis (60)	97 (58)	98 (59)	97 (58)	92 (55)
Calculation (24)	100 (24)	100 (24)	100 (24)	100 (24)
Drawing (30)	67 (20)	83 (25)	80 (24)	80 (24)

<sup>a</sup> Standard score with  $M = 10$ ,  $SD = 3$ .

<sup>b</sup> Standard score with  $M = 100$ ,  $SD = 15$ .

MTDDA = *Minnesota Test for Differential Diagnosis of Aphasia* (Schuell 1965), BDAE = *Boston Diagnostic Examination for Aphasia* (Goodglass and Kaplan 1983), ADP = *Aphasia Diagnostic Profiles* (Helm-Estabrooks 1992), WAB = *Western Aphasia Battery* (Kertesz 1982).

improvements of targeted language functions: McNeil and colleagues reported improved word-finding with a behavioural plus pharmacological treatment and Schneider and colleagues reported improved production of simple sentence structure with a verbal plus gestural treatment. Whereas both studies documented treatment effectiveness and short-term maintenance, both patients, as expected with PPA, eventually showed decline of target language behaviours. It is unclear how or if these treatments could be adapted to meet the patients' communicative needs over the progression of their disorder.

Because PPA patients may remain active and independent for years following the onset of their language symptoms, further identification and longitudinal follow-up of therapy techniques are needed. The purpose of this paper is to describe the evolving treatment regimen provided to a woman with a 4 year history of non-fluent primary progressive aphasia. Over 2.5 years of treatment at a university clinic, three different therapy approaches were used to adapt to her changing communicative abilities and needs: stimulation-facilitation, drawing, and functional communication programmes. These approaches and their associated effects on her communication profile are reported below.

### Case study

D.D. is a right-handed, English speaking, homemaker and high-school graduate. In March 1993 at the age of 64, she first reported a 'stuttering' problem to her neurologist. At that time, her medical and neurological examinations were normal, including negative CT and MRI scans, and she was subsequently diagnosed with conversion disorder.<sup>1</sup> Her communication problem continued to worsen and, in April 1994, she was referred by her neurologist for an initial speech-language assessment at Indiana University's Speech and Hearing Clinic. D.D. reported that her speech was becoming increasingly 'slurred'. She also stated that she was having word-finding difficulties in that 'I can see the words in my mind, but I can't say them'. Administration of the *Minnesota Test for Differential Diagnosis of Aphasia* (Schuell 1965) indicated non-fluent aphasia and the following communication profile (see table 1):

- (1) Mild to moderate auditory and reading comprehension impairments with difficulty processing paragraph-length material (e.g. 50% accuracy for the Reading Comprehension, Paragraph subtest);
- (2) Moderate verbal expression impairments characterized by repetition difficulties (e.g. 60% accuracy for the Repeating Phrases subtest), phonemic paraphasias (e.g. D.D. labelled a picture of a calendar as 'candler'), decreased sentence length (e.g. for the Describing Picture subtest, D.D.'s verbal output was limited to 'I see the dog and boy. Tree. Man and kite. I see the house and duck and pond.') and decreased grammatical accuracy and complexity (e.g. when given the word 'coat' and asked to produce a sentence for the Producing Sentences subtest, D.D. stated 'In the winter we get a coat wear.');

<sup>1</sup> Conversion disorder is a psychiatric illness in which there is 'an alteration in function suggestive of an organic process, but in the absence of an organic explanation...attributed to an underlying psychological conflict or need' (Philbrick *et al.* 1994, p. 138). Similar to D.D.'s case, the initial symptoms of other PPA patients have been at least temporarily attributed to a psychiatric rather than organic disorder (Hart *et al.* 1997, Kesler *et al.* 1995).

- (3) Mild written expression impairments which paralleled verbal expression deficits (e.g. for the Writing Sentences to Dictation subtest, D.D. wrote 'We have new car' for 'We have a new car').

Results of D.D.'s oral motor speech examination indicated adequate oral mechanism structure and function, and appropriate diadochokinetic rates.

D.D.'s past medical history was unremarkable except for hypertension, which was well controlled with atenolol (Tenormin®) and triamterene with thiazides (Dyazide®). She and her spouse reported no history of alcohol or substance abuse, or prior mental health treatment. There is a possible family history of dementia. D.D. reported that her mother and grandfather suffered from dementia, although no formal diagnosis was given to these individuals.

Neuropsychological testing was completed in July 1995 and revealed low average intellectual functioning with preservation of many cognitive features including orientation, gnosis, visuoperception, construction, and visual memory (see table 2). D.D. performed most poorly on those subtests which required verbal responses (e.g. WAIS-R Digit Span). Memory tests on which she was allowed to write or that required recall of non-verbal information were less impaired or within normal range (e.g. WAIS-R Information, WMS-R Visual Reproduction). Her manual motor skills were intact for speed, strength, and dexterity. A PET scan was also taken at this time and showed mild diffuse left hemisphere hypoperfusion with most marked reduction in the left frontal parietal region. Based on these assessment results, D.D.'s 2 year history of language decline, and her continued independence in activities of daily living, her diagnosis was changed at this time to primary progressive aphasia (PPA). Likewise, D.D.'s cognitive, communicative, and behavioural profile met the PPA diagnostic criteria outlined by Weintraub *et al.* (1990).

D.D. has not received in depth follow-up neuropsychological testing for several reasons. First, several researchers have questioned the validity of testing the cognitive abilities of aphasic adults given that their communication deficits may directly influence their performance on tests designed to assess functions other than language (Duffy and Petersen 1992, Hart *et al.* 1997, Helm-Estabrooks and Albert 1991); for example, it is difficult to determine whether D.D.'s poor performance of the Digit Span subtest of the WAIS-R (i.e. an age-scaled score of 3 or 5/14 correct) reflected short-term memory deficits or verbal repetition deficits (see table 2). As D.D.'s expressive and receptive language skills deteriorated over time, valid interpretation of neuropsychological findings seemed less likely, and therefore comprehensive neuropsychological testing appeared less appropriate or valuable. Second, local neuropsychological services or test materials were not available; additional testing would have required numerous trips to another community and thus additional travel time and expenses for D.D. and her spouse. Third, D.D. has indicated a dislike for formal testing, especially when she is unfamiliar with the examiner. Given these concerns and methodological restrictions, long-term monitoring of D.D.'s cognitive status has been limited to self and spousal feedback, results of certain aphasia battery subtests (e.g. Calculation and Drawing subtests of the *Western Aphasia Battery*), and documentation of activities of daily living. To date, neither D.D. nor her spouse has reported any concerns related to D.D.'s memory, attention, or problem solving skills. As shown in table 1, her calculation and drawing performances have remained stable. Furthermore, D.D.

Table 2. Results of neuropsychological testing for subject D.D.

Measure	Test score at time of measurement		
	July 1995	Sept 1995	August 1996
<i>WAIS-R</i>			
Verbal IQ	74		
Performance IQ	93 <sup>c</sup>		
Full scale IQ	82		
Information (age-scaled scores <sup>a</sup> )	8 <sup>c, d</sup>		
Digit span	3		
Arithmetic	7		
Similarities	7 <sup>d</sup>		
Picture completion	10 <sup>c</sup>		
Block design	11 <sup>c</sup>		
Digit symbol	11 <sup>c</sup>		
<i>WMS-R</i>			
Logical memory immediate (24)	12 <sup>d</sup>		
Logical memory delayed (24)	12 <sup>c, d</sup>		
Visual reproduction immediate (41)	34 <sup>c</sup>		37 <sup>c</sup>
Visual reproduction delayed (41)	33 <sup>c</sup>		37 <sup>c</sup>
Visuospatial			
Judgment of line orientation (30)	24 <sup>c</sup>		
Facial recognition (54)	51 <sup>c</sup>		
Trail making			
Part A	66 s		69 s
Part B	175 s		
<i>WRAT-R</i> (Standard scores <sup>b</sup> )			
Reading	66		
Spelling	78		
Arithmetic	106 <sup>c</sup>		
<i>DRS</i>			
Total score (144)		115	
Attention (37)		34	
Initiation and perseveration (37)		27	
Construction (6)		6 <sup>c</sup>	6 <sup>c</sup>
Conceptualization (39)		26	
Memory (25)		22	

<sup>a</sup> Mean = 10, SD = 3.

<sup>b</sup> Mean = 100, SD = 15.

<sup>c</sup> Test or subtests on which D.D. scored at or above average.

<sup>d</sup> Subtests on which D.D. was allowed to write her responses.

WAIS-R = *Wechsler Adult Intelligence Scales—Revised* (Wechsler 1981), WMS-R = *Wechsler Memory Scale—Revised* (Wechsler 1987), WRAT-R = *Wide Range Achievement Test—Revised* (Jastak and Wilkinson 1984), DRS = *Dementia Rating Scale* (Mattis 1988). Figures in parentheses indicate maximum scores.

has remained active and independent over the 4 years since the onset of her communication problems. She continues to complete household responsibilities and daily activities include shopping, cooking, driving, and banking, and she and her husband regularly fish, hike, and travel.

Recently, the investigator administered a small battery of neuropsychological tests to D.D. (see table 2); these tests were chosen because of their availability and their minimal language requirements, and because previous results were available

Table 3. Results of functional communication assessments

Measure	Test score at time of measurement (%)			
	Sept 1995	Dec 1995	May 1996	Dec 1996
CADL (146)	83 % (121)		90 % (131)	88 % (128)
CETI (100 mm)				
Spouse rating D.D.		55.4	44.8	60.8
D.D. self-rating			41.1	54.6
ASHA FACS				
Communication independence (max. 7)				6
Social communication				5.4
Basic needs				7.0
Reading, writing, numbers				7.0
Daily planning				7.0
Qualitative dimensions (max. 5)				4.1
Adequacy				4.0
Appropriateness				4.5
Promptness				4.3
Communication sharing				3.5

CADL = *Communicative Abilities in Daily Living* (Holland 1980), CETI = *Communicative Effectiveness Index* (Lomas *et al.* 1989), ASHA FACS = *American Speech-Language-Hearing Association Functional Assessment of Communication Skills for Adults* (Frattali *et al.* 1995). Raw scores and maximal scores are provided in parentheses.

for direct comparison. D.D. showed minimal or no change in her performance of the WMS-R Visual Reproduction Immediate and Delayed subtests, Trail Making Test Part A, or the Construction items of the DRS. For example, her performance of the Visual Reproduction subtests indicated that she continues to display above average visual memory skills. Interestingly, this subtest is considered to be very sensitive to the cognitive decline of adults with dementia, both in the early and late stages of the disease (Kaszniak *et al.* 1978, Mitrushina *et al.* 1988). Therefore, these recent neuropsychological findings continue to support the validity of D.D.'s PPA diagnosis.

Since July 1995, D.D. has received pharmacologic treatment for depression (Paxil) and nervousness (Klonopin); she has stated that this depression is primarily related to her communication problems. She and her spouse have reported no other psychological problems and administration of the Frontal Lobe Personality Scale (Grace *et al.* 1998, Paulsen *et al.* 1996) in September 1995 was unremarkable for disinhibition, apathy, or executive dysfunction.

Longitudinal speech-language testing has indicated progressive decline in most modalities (see table 1). D.D.'s verbal expression has been most severely affected and is characterized by increasing agrammatism, anomia, phonemic paraphasias, and apraxia of speech. More recently, D.D. has also shown decreases in her auditory comprehension abilities. Although impaired, reading and writing have remained modalities of relative strength. Results of functional communication assessments (e.g. CADL, ASHA FACS) indicate that despite progressive deterioration of her linguistic skills, D.D. remains a fairly successful, although somewhat passive, communicator (see table 3). This profile (i.e. good communication despite poor language skills) suggests that D.D., like adults with aphasia due to stroke, has relatively preserved communicative competence and has

been able to compensate for her linguistic inadequacies (Busch *et al.* 1989, Herrman *et al.* 1988, Holland 1996). Audiological evaluation in June 1996 showed bilateral borderline-normal hearing up to 3 kHz, sloping to moderate sensorineural loss at higher frequencies. Further details regarding changes in D.D.'s communication skills over time are provided in each of the following treatment sections.

### Treatment 1 (June 1994 to April 1995)

The first treatment approach occurred prior to the investigator's direct involvement with D.D.; therefore, the following description of procedures and data was based entirely on the clinical reports of graduate student clinicians and patient and spousal report. It should also be acknowledged that D.D. did not receive a diagnosis of PPA until the summer following this treatment phase.

Initial language therapy consisted of traditional stimulus-response activities which were designed to stimulate and facilitate D.D.'s auditory and reading comprehension skills at the paragraph level and her spoken and written expression at the three- to four-word sentence level (see table 4). D.D. either listened to or read a paragraph that was at least five sentences in length, then answered yes-no inferencing questions and open-ended questions that required one- to two-word responses; the length of the paragraphs that she was required to read and the number of sentences that she was required to answer varied slightly across student clinicians and treatment semesters. When D.D. erred, she or the clinician, the latter if it was an auditory comprehension task, reread the one or two sentences that pertained to the given question in order to highlight the relevant information. During transcription activities, D.D.'s errors were identified and she was then required to rewrite the stimulus. During spoken and written sentence production activities, D.D. described action pictures using the sentence structure specified by the clinician (e.g. *subject is/are verb-ing*). She was again provided feedback concerning the loci of her errors, then required to reproduce the target sentence structure. Towards the end of this treatment phase, D.D. was encouraged to use sentences for a set period of conversation; however, data concerning this therapy activity were not available. Individual 45 minute sessions were provided twice weekly for a total 46.5 h of therapy.

Table 4 shows D.D.'s performance of pre- and post-treatment probes (i.e. her performance of each target task during a single pre- or post-treatment probing session). These data indicated that D.D. progressed and achieved 80 % or greater accuracy on all tasks following each treatment period. Following no treatment phases, D.D. displayed partial or complete return of targeted language behaviours to baseline levels. She also demonstrated improved performance of treated behaviours despite simultaneous decline of other language functions such as repetition (i.e. dropping from 60 % in April 1994 to 48 % in December 1994 to 0 % in January 1995), or naming (i.e., dropping from 95 % in April 1994 to 88 % in December 1994 to 72 % in January 1995) (see table 1). These observations suggested that D.D.'s improvement was related to treatment.

Whereas these data indicated that D.D. was benefiting from treatment, informal observations signified inadequacy in the current therapy approach. For example, little carry over of treatment effects to D.D.'s everyday communication interactions was reported by D.D. or her spouse. Furthermore, treatment was not offsetting the progressive deterioration of her spoken discourse skills. Throughout this treatment



Table 5. D.D.'s pre- and post-treatment (Back-to-the-Drawing Board) performances

	Pre-Tx, M Sept 1995	Oct 2	Oct 18	Nov 8	Nov 27	Post-Tx, M Dec 1995
Drawing measures						
Correctly identified						
Main events	0.44					0.63
	0.70					1.15*
	1.11					1.26
Detail rating	39.8					40.5
	44.3					60.1*
	42.3					49.5
Clarity rating	51.0					53.0
	44.6					58.4*
	41.9					50.2
Control measures						
Convergent naming	75	70	80	80	80	75
	15	14	16	16	16	15
Divergent naming	9	10	9	11	9	11
	Max. no of responses					
Grammatical completion (TOLD-P2)	6					6
	Correct (30)					15
	Omissions					7
	Substitution					2
	No response					
Generalization measures						
PACE task	2.4					3.3
Unfamiliar partner	4.8					3.2
	1					0
PACE task	2.0					2.4
Familiar partner	5.0					3.6
	0					1
	Drawn cues					

\* Significantly higher than pre-treatment performance ( $p < 0.05$ ).

TOLD-P2 = Test of Language Development—Primary 2 (Newcomer and Hammill 1988), CRS = Communicative Rating Scale (Davis and Wilcox 1981).

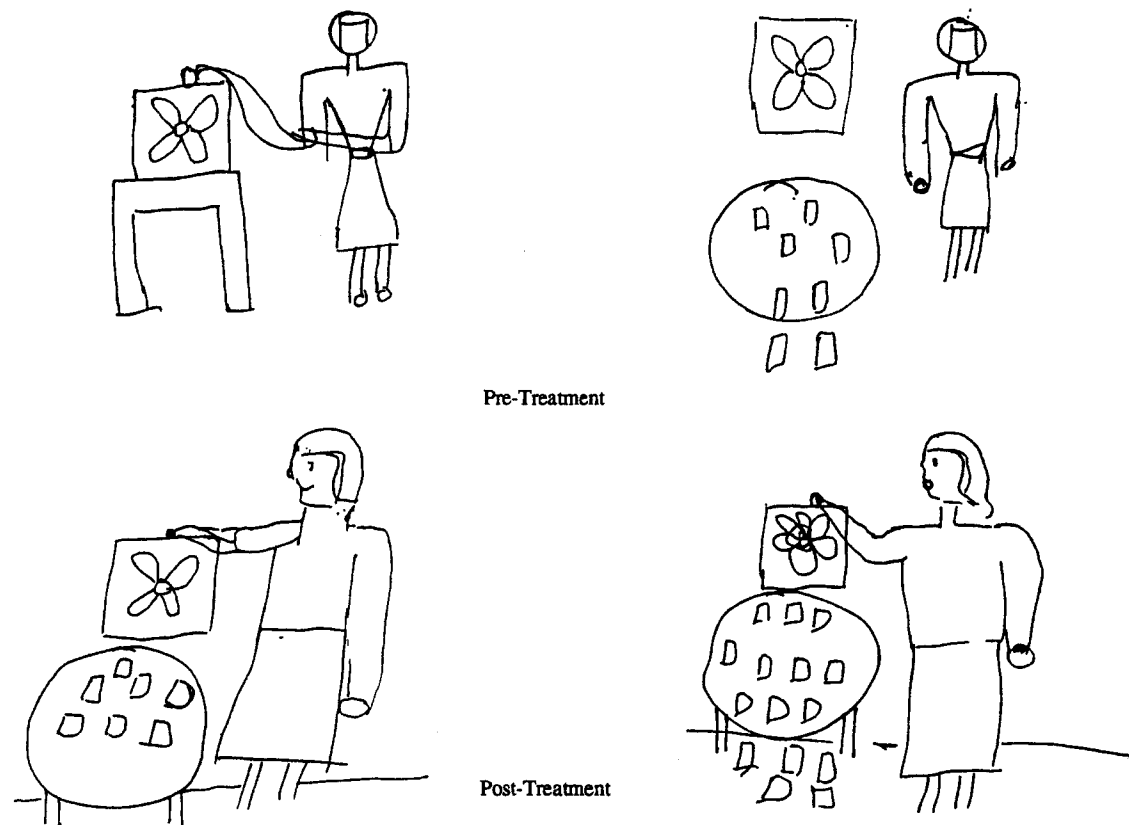


Figure 1. An example of D.D.'s pre- and post-treatment drawings of a two-part scene from the Daily Mishaps Test (Helm-Estabrooks and Albert 1991). As described by these researchers (p. 196), the scene is to depict the following: 'A woman turns on a fan. As the fan rotates, the papers on a desk blow around'. Group mean ratings for this pre-treatment drawing were 61.8/100 mm for details, 66.4/100 mm for clarity, and 1.1/2 for number of main ideas correctly identified. Group mean ratings for this post-treatment drawing were 72.0/100 mm for details, 73.5/100 mm for clarity, and 1.6/2 for number of main ideas correctly identified.

were necessary to improve the given drawing. The clarity scale was used to assess drawing quality and precision (e.g. straight lines, absence of overlapping images) and meaningfulness (e.g. main idea transparency).

Table 5 shows that D.D.'s post-treatment drawings depicted main events more accurately and were judged to have greater detail and clarity than her pre-treatment drawings. (See figure 1 for an example of D.D.'s pre- and post-treatment DMT drawings.) Results from pairwise *t*-tests indicated that the difference between pre- and post-treatment drawings was significant ( $p < 0.05$ ) for only drawings of two-part scenes (correctly identified main events  $t = 3.229$ ,  $p = 0.004$ , drawing detail  $t = 3.304$ ,  $p = 0.003$ , drawing clarity  $t = 3.041$ ,  $p = 0.006$ ). D.D. also showed better performance of the WAB Drawing subtest, improving from 67% to 83% (see table 1). Treatment effects were further supported by a constant trend on control tests (see table 5): D.D. demonstrated no change in her performance of the Grammatical Completion subtest of the TOLD-P2 (Newcomer and Hammill 1988), or in her convergent and divergent naming skills as measured by a 20 item phrase completion task (Murray 1996) and a word fluency measure (i.e. producing the names of as many vegetables as possible in 1 min), respectively. Lastly, D.D. and her spouse reported that she enjoyed the drawing therapy programme, especially in relation to previous therapy tasks. D.D.'s affinity for the drawing

programme might be related to the fact that it took the focus of therapy away from her verbal expression skills, a communication modality for which she reported frustration and embarrassment.

Despite positive treatment effects, D.D. showed little generalization of improved drawing performance. For example, although she was encouraged to use any modality when completing control tests of convergent and divergent naming, all of her responses were written. PACE tasks were also used to examine for generalization (see table 5). D.D. was required to communicate the names of two sets (i.e. five items per set) of famous people (e.g. Elvis Presley), events and special occasions (e.g. St Valentine's day), and places (e.g. Statue of Liberty) using cues in any modality to a familiar interactant (her spouse) and to an unfamiliar interactant (a graduate student). Each set of names was communicated to each interactant in the opposite presentation order (i.e. during the post-treatment assessment, D.D. communicated to her spouse the same set of names that she communicated to the graduate student during the pre-treatment assessment). D.D.'s messages were scored by two raters using the four-point Communicative Rating Scale developed by Davis and Wilcox (1981); inter-rater agreement was 100 %. Although there was slight improvement in D.D.'s communication efficiency (i.e. higher Communicative Rating Scale ratings, fewer cues given by D.D.), there was no increase in her use of drawing during the PACE activity. D.D. and her spouse also reported little spontaneous use of drawing in her communication interactions outside of the therapy room.

It is possible that task or stimuli constraints, at least in part, were responsible for D.D.'s minimal use of drawing during post-treatment testing; that is, the concepts that she was required to communicate during these tasks were relatively simple and short and consequently were within her written language capabilities. For example, the divergent naming task required listing associates within a single category; given the 1 minute time constraint for this task, the most efficient strategy would be to exploit first one's written vocabulary prior to drawing associates for which one is having lexical or graphemic retrieval difficulties. Likewise, the stimuli that she was required to communicate during the PACE activity could easily be communicated using short written word cues; that is, writing 'Feb 14', as D.D. did, is a quicker and more concise clue than drawing a heart (which could signify 'love', 'valentine', 'heart', etc.).

Poor generalization to settings outside of the clinic may have been partially due to the minimal training D.D.'s spouse received. Following BDB training, Helm-Estabrooks and Albert (1991) recommend therapy sessions to train communication partners. During these sessions, communication partners are taught questioning strategies that facilitate extraction of information from the aphasic adult's drawings. D.D.'s spouse received only two of these sessions, which may have been insufficient exposure to these questioning strategies and thus insufficient development of his skills to encourage and facilitate D.D.'s use of drawing at home. Further post-BDB training was not provided because during these two spouse-patient sessions, many negative interaction characteristics were observed (e.g. D.D.'s spouse would correct her spelling errors rather than acknowledge or comment on her message). Therefore it was decided that during D.D.'s next semester of therapy, addressing these interaction issues was a treatment priority as opposed to providing further drawing training.

### Treatment 3 (January 1996 to present)

Following BDB training in December 1995, D.D.'s communication skills were re-evaluated. Findings from the ADP (Helm-Estabrooks and Albert 1991) and the WAB (Kertesz 1982) indicated that her language skills in all modalities showed little change compared to findings from her September 1995 evaluation (see table 1). At this time, in depth assessment of her verbal expression skills was also completed in an attempt to differentiate motor speech deficits from expressive language deficits. Results from Dabul's (1979) *Apraxia Battery for Adults* indicated that D.D. presented with no limb or oral apraxia but with severe to profound apraxia of speech. Apraxic symptoms included articulatory groping, vowel distortions, difficulty self-correcting, inconsistent errors, and increased frequency of articulatory errors as word length increased. Orofacial structure, oral-motor performance, and respiration appeared normal during an oral mechanism examination; reductions in vocal loudness, pitch and loudness variability, and speaking rate were noted. The *Photo Articulation Test* (Pendergast *et al.* 1969) was given to assess D.D.'s speech sound production skills. D.D. made 31 errors (out of a possible 94) which involved single consonant, consonant cluster, and vowel and diphthong productions, and which primarily occurred in word medial and final positions. Error types included cluster reductions (e.g. /sar/ for 'stars'), stopping (e.g. /tit/ for 'teeth'), deaffrication (e.g. /sar/ for 'chair'), and devoicing (e.g. /kis/ for 'keys'). To examine intelligibility, the *Assessment of Intelligibility of Dysarthric Speech* (Yorkston and Beukelman 1981) was administered. Using the multiple choice format, D.D.'s single word intelligibility was 46 % (i.e. 23 out of 50 words were correctly identified by a naive listener). At the sentence level, D.D.'s speech was only 20 % intelligible (i.e. 43 out of 220 words were correctly transcribed by a naive listener). Collectively, these findings indicated that although D.D. unquestionably presented with aphasia, motor speech deficits were also contributing to her reduced verbal output. The co-existence of both apraxia of speech and non-fluent aphasia has been reported previously in several PPA case studies (Ackermann *et al.* 1997, Hart *et al.* 1997, Rogers and Alarcon 1997).

To target the frustration and the frequent communication breakdowns demonstrated and reported by D.D. and her spouse, a more pragmatic or functional communication approach was adopted to encourage multimodality communication and flexibility and to facilitate communicative interactions (Hickey *et al.* 1996, Kagan and Gailey 1993, Simmons *et al.* 1987). During this treatment phase, D.D. attended individual as well as group treatment sessions and by December 1996, she had completed 41 hours of individual therapy and 10 hours of aphasia support group.

Individual treatment focused on improving communication interactions between D.D. and her spouse. The first step was to require D.D.'s husband to observe or participate in all of her individual therapy sessions. Next, the clinician identified for D.D. and her spouse which turn-taking and repair skills they were using (based on pre-treatment conversational samples) and discussed the effectiveness of their strategies as well as possible alternatives. Role-playing, PACE tasks, and videotape review were then used to model and elicit specific strategies designed to facilitate interactions (e.g. encourage D.D.'s use of assertive conversational behaviours such as topic initiation or requests for information) and repair conversational breakdowns (e.g. encourage D.D.'s spouse to request elaboration rather than

repetition of unclear messages). Role-playing sessions were typically completed by D.D. and her clinician. For example, D.D. (the customer) and the clinician (the waitress) would pretend to be at a restaurant; the clinician would create communication breakdowns or problems (e.g. mix up D.D.'s order) so that D.D. could practice identifying and repairing communication breakdowns and so that the clinician could model appropriate repair strategies for D.D.'s spouse. During PACE tasks, D.D. and her husband were provided with specific message stimuli to convey to each other (e.g. recent news events, famous people or places, personal information and events). They were required to communicate these specific messages to their partner as quickly as possible, using any communication modality they desired but without telling their partner the exact word on the card. This activity was designed to increase D.D.'s use of requests for information and to encourage the couple's use of multimodality communication and repair strategies. D.D. and her spouse also reviewed videotaped samples of their conversations; samples were taken from their conversations within the therapy room during which they discussed two to three topics provided by the clinician or chosen by themselves. When viewing the samples, D.D. and her spouse referred to a written list of conversational features and strategies to identify which ones they had used. The clinician and the couple then discussed positive and negative aspects of the conversational interaction.

At the beginning of this treatment phase, D.D. and her spouse requested information on the appropriateness and feasibility of an electronic augmentative communication device. Based on D.D.'s cognitive, linguistic, perceptual, and physical capabilities, her communicative needs (e.g. make personal phone calls, communicate with grandchildren who were not yet able to read her written messages), and a successful trial period, a Sentient Systems' DynaVox was selected. Because of safety issues (i.e. D.D. needed a means to make emergency phone calls if she was home alone), the family was able to obtain funding of this device through their supplemental medical insurance.

While D.D. awaited funding approval and the arrival of her own device, she, her daughter-in-law (who became responsible for all device programming), her spouse, and the clinician assembled functional vocabulary words and phrases and arranged them into categories (which would later become DynaVox 'pages'). Categories/pages included people (e.g. names of family members and friends), places (e.g. locations that she frequents on a daily or weekly basis), time (e.g. days of the week, months, time of day), activities (e.g. daily activities and hobbies), card playing (e.g. 'queen', 'spades'), feelings, and special pages for making telephone calls (e.g. 'Please call back later') and small talk (e.g. 'How are you today?'). Symbols for messages were then selected from the DynaVox user manual and copied for use on communication boards. Placement of symbols was conjointly decided by D.D. and her clinician. These communication boards were used to familiarize D.D. with the symbols and their locations prior to delivery of her actual device.

Once D.D. received her own DynaVox, it was programmed with the pre-selected vocabulary. Page and vocabulary additions and amendments continue to be made as needs or problems arise; for example, a present programming goal is to add the narration of a children's book so that D.D. can tell her grandchildren a story. A variety of activities have been used to foster D.D.'s use of this augmentative device. Drill activities entailed repetitively asking D.D. open-ended questions (e.g. 'What is your name?'; 'What did you eat for breakfast?'). These



tasks were utilized to increase D.D.'s symbol selection speed and accuracy. It should be noted that symbol sequencing was never directly trained because D.D. spontaneously combined and sequenced symbols. D.D. and her clinician also completed role-playing activities. Situations such as making a doctor's appointment, ordering food at a restaurant, or phoning a friend, were acted out in person and using a speaker phone to encourage D.D.'s use of the device during both face-to-face conversations as well as telephone conversations. Lastly, each therapy session included some conversation between D.D. and a familiar (e.g. her spouse, the clinician) or unfamiliar (e.g. another graduate student) communication partner. These conversations were completed to encourage D.D.'s use of the DynaVox as well as to target the previously described communication interaction goals. For example, D.D. was encouraged to use a variety of modalities during these interactions (i.e. not just DynaVox use, but also drawing, writing, and gesturing), and in addition to the previously identified conversational repair strategies, she was taught repair strategies specific to communication with the DynaVox (e.g. if the listener did not understand D.D.'s message via the verbal output of the DynaVox, she was taught to show the listener the machine's written display of her message).

During this treatment phase, D.D. and her spouse also joined the Indiana University Aphasia Support Group. D.D. attended group therapy with another woman who presented with severe non-fluent aphasia as a consequence of a left hemisphere stroke. This woman's verbal output was also severely limited and was restricted to one recurrent utterance (i.e. 'Oh my'). Group activities were designed to encourage D.D.'s generalization of repair strategies, assertive conversational behaviours, and multimodality communication including DynaVox use. D.D.'s spouse attended the caregivers' group which provided social-emotional support and education, as well as PACE and role-playing activities to target communication facilitation and repair strategies.

Qualitative analysis was conducted on pre- and post-treatment conversational interactions between D.D. and her spouse. Conversations were elicited by encouraging the couple to discuss two to three topics of the clinician's or their choice; their conversations averaged 8 minutes in length. Table 6 shows that D.D. and her spouse increased their repertoire and use of repair strategies (see Appendix for a description of discourse categories). For example, during initial communication interactions, the predominant repair strategy used and requested by both D.D. and her spouse was direct repetition of a message, usually resulting in numerous repetitions of the same statement or question, and frustration on the part of the message sender and recipient. By December 1996, D.D.'s spouse was observed to encourage her elaboration of messages and also asked her to use a different communication modality. D.D. also began to demonstrate more active participation in conversational interactions. Although overall she continues to be a passive communicator, during a post-treatment conversation with an unfamiliar partner at least one occurrence of topic initiation (e.g. using the DynaVox she asked the clinician 'Thanksgiving how?') and one query or request for information were observed. Both D.D. and her spouse also exploited a larger number of communication modalities during final conversational samples; interestingly, 4 months following Back-to-the-Drawing-Board therapy, D.D. spontaneously began to use drawing to augment her written messages.

Table 7 shows that D.D. quickly adapted to her DynaVox and demonstrated

Table 7. Summary of D.D.'s DynaVox performance

Activity	Measure	Feb* 1996	Sept 1996	Dec 1996
Vocabulary	Pages	6		12
	Symbols	30		≈ 200
Drill practice (e.g. answering open ended questions, 'Where do you live?')	Mean accuracy (%)		80	95
	Mean latency <sup>b</sup> (s)		15	5
Situational practice (e.g. phone calls, PACE task, conversation)	Mean accuracy (%)	80	77	90
	Latency range (s)	2-31	1-30	1-26
	Max. number of symbols/turn	4	3	6

\* Data for this time period were based upon D.D.'s final performance following a 2 week trial period with a rented DynaVox. Several other augmentative devices were tried during the spring 1996 treatment semester as well. D.D. did not purchase her own DynaVox until July 1996 (due to insurance delays) and formal training with the device did not resume until September 1996.

<sup>b</sup> Latency figures reflect the length of time D.D. required to select a single symbol, as opposed to a complete message.

improved message accuracy and length and response latency. The family also purchased a speaker telephone to allow D.D.'s use of the device to make phone calls at home. Unfortunately, D.D. and her spouse have reported minimal use of the DynaVox outside of the home or clinic. Poor generalization may be, at least in part, due to the DynaVox's size and consequently, due to portability problems. Efforts are now directed towards trading in D.D.'s DynaVox for Sentient Systems' newer and smaller DynaMite in an attempt to increase the number of communicative settings in which she can use her augmentative system.

Despite D.D.'s different aphasia aetiology (i.e. progressive vs. static lesion), she and her spouse reported that they enjoyed and valued the informational and emotional support provided by their weekly aphasia and spouse groups, respectively. During her group sessions, both D.D. and her fellow group member used a variety of communication modalities. D.D. was an active conversational participant; she frequently initiated conversation via writing and used many of the repair strategies practised during her individual therapy sessions.

The results of post-treatment functional communication assessments also supported positive qualitative and quantitative findings (see table 3). It is speculated that D.D.'s improved or maintained performance on the CADL (Holland 1980) and the CETI (Lomas *et al.* 1989) and her relatively high ratings on the ASHA FACS (Frattali *et al.* 1995) suggest that treatment, at least in part, helped D.D. retain relatively effective and independent functional communication, despite the progressive decline of her linguistic abilities.

### Summary

Whereas previous studies of speech-language treatment for primary progressive aphasia have focused on a single therapy approach, the present case report described a dynamic intervention programme in which treatment proceeded from a stimulation-facilitation approach to a drawing programme and lastly, to a dyad-



oriented functional communication approach. Quantitative and qualitative findings indicated that D.D. displayed improvements during each of these treatment approaches.

Similar to other PPA patients in the initial years of their clinical syndrome (McNeil *et al.* 1995, Northen *et al.* 1990), D.D. showed improved performance of target language behaviours following stimulation-facilitation treatment but failed to generalize improvements to daily discourse contexts. Next, back-to-the-drawing-board training (Morgan and Helm-Estabrooks 1987) was implemented and D.D. showed improved drawing performance on post-treatment measures. Although minimal generalization of drawing to everyday communication interactions was initially noted, 4 months post-treatment D.D. spontaneously began to augment her written messages with drawing. Lastly, a dyadic, functional communication approach was adopted, and an electronic augmentative device, the DynaVox, was introduced. Qualitative analysis of post-treatment interactions indicated that both D.D. and her spouse benefited from dyad-oriented therapy activities (Simmons *et al.* 1987) and increased their range of strategies to circumvent and prevent communication breakdowns. D.D. also showed successful acquisition and use of the DynaVox; provision of other augmentative devices such as communication boards and books has been previously described in the PPA literature (Béland and Ska 1992, Kesler *et al.* 1995, Northen *et al.* 1990). Future rehabilitation efforts will continue to adapt to the changing needs of D.D. and her family. The goal of this treatment will be to maintain effective communication between D.D. and her loved ones for as long as possible.

In conclusion, professionals such as neurologists, neuropsychologists, and speech-language pathologists are encouraged to advocate for the provision of services to patients with PPA. Findings from D.D.'s longitudinal treatment regimen support previous reports of treatment effectiveness in PPA (McNeil *et al.* 1995, Schneider *et al.* 1996) and suggest that long-term speech-language treatment of PPA is appropriate despite the traditional therapeutic nihilism that frequently accompanies diagnosis of progressive disorders (Clarke 1995). PPA patients, particularly those with the non-fluent variant (Duffy and Petersen 1992, Rogers and Alarcon 1997, Watt *et al.* 1997), may suffer isolated language decline for years prior to the onset of other cognitive impairments and therefore remain viable candidates for many of the treatment methods used to treat traditional aphasic patients with 'static' lesions.

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### Appendix: Discourse categories

Category	Behavioural definition
Request repetition	Message recipient requests that sender repeat his or her message.
Response repetition	Sender repeats his or her message spontaneously or upon request to repair an unsuccessful message transmission.
Request clarification (Y/N)	Message recipient requests a clarification in the form of a yes/no question when he or she did not completely understand the sender's message (e.g. 'Do you mean you want to go hiking tomorrow?').
Response elaboration	Sender elaborates his or her message (provides more information) to repair an unsuccessful message transmission.
Request elaboration	Message recipient asks for more information when he or she did not completely understand the sender's message (e.g. 'When do you want to go hiking?').
Show DynaVox screen	D.D. shows the written form of her message on the DynaVox screen when the message recipient does not appear to understand the machine's verbal output.
Encourage multimodality communication	Message recipient requests that the sender exploit additional communication modalities when he or she did not completely understand the sender's message (e.g. 'Try drawing a picture').
Response revision	Sender revises his or her message to repair an unsuccessful message transmission. No new information is provided but rather the form of the message is changed or simplified.
Turn-taking	Communication interactant displays appropriate turn-taking skills (e.g. responding when spoken to, indicating the end of their conversational turn, waiting for communication partner to complete his or her turn).
Topic maintenance	Communication interactant maintains topic of conversation (operationalized as at least two turns for this study).
Topic shift	Communication interactant is able to follow a shift in conversational topic (e.g. does not perseverate on prior conversational topic).
Topic initiation	Communication interactant initiates a new topic.
Query	Communication interactant asks a question regarding the given conversational topic. For this study, requests for repetition, clarification, or elaboration were not considered queries.