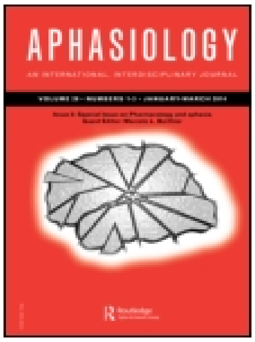

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Analysing speech problems in a longitudinal case study of logopenic variant PPA

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Background: Primary progressive aphasia (PPA) is a neurodegenerative form of dementia in which gradually worsening language impairments are the prominent feature in the initial stages. PPA is commonly differentiated into three variants: nonfluent agrammatic (PPA-NVF), semantic (PPA-SV), and logopenic (PPA-LV).
Aims: This article provides a longitudinal description of changes in picture description produced by a woman with PPA-LV, introduces a reliable new measure that captures those changes, and relates the measured changes to raters' perceptions of changes in discourse quality.
Method & Procedures: Seven oral descriptions of the Boston Diagnostic Aphasia Examination (BDAE) Cookie Theft picture were digitally recorded over the course of 27 months and later transcribed. Transcriptions were analysed using a new adaptation of the Linguistic Communication Measure (LCM) and the Linguistic Communication Measure-Revised Cantonese (LCM-RC) designed to be sensitive to the features of PPA-LV. We have named this third form the LCM, the Linguistic Communication Measure–-Speech Sounds (LCM-SS). Audio recordings of the seven picture descriptions plus three produced by typical speakers of similar age were rated for *goodness* by 15 raters.
Outcomes & Results: Goodness ratings of the participants' speech samples decreased steadily over the 27 months. Although our previous measures of discourse quality (LCM, LCMC-RC) appeared to work well for capturing many of the speakers with vascular aphasia, they failed to capture the nature of this participant's decline: Her lexical access slowed over time, but did not become more error-prone, and morphosyntactic components did not worsen, with errors remaining low to almost absent. However, speech sound errors and repetitions increased steadily over the 27 months. The new measure, LCM-SS, succeeded in capturing this pattern of decline: Several of the LCM-SS measures were highly correlated to ratings of *goodness*, and two of the LCM-SS indices (sound errors and grammatical errors) accounted for 98% of the variance in the goodness ratings.
Conclusions: Over the course of 27 months, the most significant change in this participant's Cookie Theft descriptions was the steady increase in sound errors, in the context of decreased efficiency in lexical retrieval and relatively stable grammatical form. This pattern was also highly related to listeners' perceptions of the quality of discourse. Neither of the previous versions of the LCM captured this debilitating increase in

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sound errors, but adding the index of sound errors to those previous versions resulted in an analysis method that was sensitive to the linguistic features exhibited by this participant with PPA-LV.

Keywords: Primary progressive aphasia; Logopenic; Speech sound errors; Analysis of connected speech; Assessment; Diagnosis; Rater perception.

Primary progressive aphasia (PPA) is a neurodegenerative form of dementia in which gradually worsening language impairments associated with atrophy of left frontal and temporal regions are the prominent initial features (Gorno-Tempini et al., 2004; Mesulam, 1982, 2001, 2003). Until recently, the most common differentiation of PPA patterns has been either semantic dementia vs. progressive nonfluent aphasia, or fluent vs. nonfluent. But this taxonomy proved to be ineffective in classifying a third group that Gorno-Tempini et al. (2004) described as having primary progressive aphasia logopenic variant (PPA-LV). Consequently, Gorno-Tempini et al. (2011) proposed a common framework for classifying PPA into three variants: nonfluent/agrammatic, semantic, and logopenic.

Once the diagnostic criteria of PPA have been met, careful evaluation of speech and language features is needed to arrive at a clinical classification of the type of PPA (Figure 1). Differential diagnosis of PPA and its variants requires "multiple sources of information including speech and language tests (including assessment of connected speech), neuropsychological testing, neurological examination and neuroimaging" (Wilson et al., 2010, p. 2085). Gorno-Tempini et al. (2011) suggest that a

All of the following must be true:

- Language difficulty is the most prominent clinical feature.
- Language impairments are the primary cause of difficulties with activities of daily living.
- Language difficulties are the most prominent deficit at onset and in initial phases.
- Word finding difficulties in spontaneous speech and confrontation naming tasks
- Impaired repetition of phrases and sentences
- The pattern of deficits cannot be better accounted for by some other disorder.
- The cognitive impairments cannot be better accounted for by a psychiatric diagnosis.
- There cannot be prominent episodic memory, visual memory and visuoperceptual impairments in the initial phases.
- There cannot be a prominent initial behavioral di sturbance.

At least three of the following must be present:

- Speech sound errors in spontaneous speech and confrontation naming
- Good single-word comprehension and object knowledge
- Good motor speech abilities
- No frank agrammatism

Figure 1. Criteria for clinical diagnosis of logopenic primary progressive aphasia (PPA; Gorno-Tempini et al., 2011).

"detailed evaluation by a speech-language pathologist" (p. 1008) will provide the most reliable source of information regarding speech and language.

Speech-language pathologists have a variety of standardised assessment tools available for evaluation of fluency, word finding, repetition, auditory comprehension (Goodglass, Kaplan, & Barresi, 2000; Kay, Coltheart, & Lesser, 1992; Kertesz, 2006) and motor speech abilities (e.g., Dabul, 2000; Enderby & Palmer, 2008). Although generally designed for evaluation of people with aphasia resulting from vascular lesions, tools like these also are used in the evaluation of language skills in people with PPA. Some of these assessments include a task in which connected speech is elicited—usually a description of connected speech. Analysis of connected speech also is thought to be the most ecologically valid indication of real-life functional language abilities, because it provides a means of observing how isolated language skills work in action (Armstrong, 2000). However, different types of discourse elicitation tasks place different demands on the speaker so that different measures of each may be called for.

In existing tools, the analysis of the discourse sample is typically designed to differentiate fluent from nonfluent aphasia, which is not a differential diagnostic feature for PPA-LV. Some researchers classified their logopenic participants as fluent; other researchers considered their logopenic participants to be nonfluent (Gorno-Tempini et al., 2008). If the Boston classification is used (Goodglass et al., 2000), the requirement to distinguish phonological errors from problems with articulatory agility increases the difficulty of determining fluency in these participants. Finally, the PPA-LV participants showed an unexpected combination of reduced phrase length with a relatively preserved diversity of grammatical forms—thoroughly confounding the fluent/nonfluent divide. In summary, simply extending speech analysis measures developed for vascular aphasia obscures essential features of PPA-LV, and a new approach is called for.

Illustrating this challenge further, in 2010, Wilson and colleagues reported on the connected speech patterns in three variants of PPA. Samples were collected in a picture description task and analysed using a procedure based on quantitative production analysis (QPA) (Berndt, Wayland, Rochon, Saffran, & Schwartz, 2000; Saffran, Sloan-Berndt, & Schwartz, 1989). Analysis of discourse produced by 11 people with PPA-LV also provided an unclear picture of fluency. The speech rate of these participants was not as fast as that of the semantic and not as slow as the nonfluent variants. Sound-level errors, if present, tended to be in the form of what appeared to be phonological paraphasia rather than distortions resulting from motor speech impairments. These participants produced more false starts, filled pauses, and repaired sequences than those with nonfluent and semantic PPA variants. Grammatical form was relatively intact but when errors occurred, they were paragrammatic rather than agrammatic. These authors concluded that a detailed multidimensional analysis of discourse gathered from a picture description task aids in the differential diagnosis of PPA variants. Existing longitudinal studies of PPA (Libon et al., 2009; Rogalski et al., 2011) describe isolated speech, language and nonlinguistic cognitive performances, but they have not examined deterioration in picture description or any other kind of connected discourse.

The primary purpose of this article is to report on the results of a detailed multidimensional analysis of connected speech gathered over the course of 27 months in a participant with PPA-LV. We developed a new discourse analysis tool that would be sensitive to observed performance patterns in this participant with logopenic PPA and that would be able to capture difficulties that were escaping existing measures—in particular, BYR's speech sound production difficulties. In addition, this article looks at the relationship between specific features of discourse production and qualitative perceptions to understand what contributed to worsening communicative success.

METHOD

Participants

Speaker with PPA-LV. BYR, the participant with PPA, was born and raised in China, speaking the Shanghainese dialect; however, she attended school where the primary language of instruction was British English. She also spoke Mandarin and Cantonese, although she said that she was most proficient with English and Shanghainese as an adult. BYR received a college degree in Ireland and used English in all of her subsequent employment, e.g., as an executive secretary in a large firm. At the time of the video recordings used in this study, she had been living in the United States approximately 40 years with her monolingual American husband. We observed that she was extremely proficient in English, retaining a slight British accent.

At around the age of 70 years, BYR began experiencing slowly worsening word-finding problems, which she reported as having found very distressing, and she was diagnosed with PPA at the age of 72 years after being referred for neurology and speech-language pathology consults by her primary care physician. At the age of 76 years, BYR began providing longitudinal data to our research team and she continued to provide data in 10 sessions at 3- to 4- month intervals over the course of 27 months. Seven of these sessions included recordings of BYR describing the Boston Diagnostic Aphasia Examination (BDAE) Cookie Theft picture (Goodglass et al., 2000), and this is the data being analysed in this report.

Initial testing by our team (Filley et al., 2006) revealed that cranial nerves, motor system, coordination, gait, and reflexes were within normal limits. BYR scored 27/30 on the Mini-Mental State Examination (MMSE; Folstein, Folstein, & McHugh, 1975) and 17/18 on the Frontal Assessment Battery (DuBois, Slachevsky, Litvan, & Pillon, 2000). Reading, writing, praxis, gnosis, calculations, right-left orientation, clock drawing, and comportment were preserved, but word-finding pauses and sound-level errors were evident in both English and Shanghainese. Because no weakness of speech musculature, reduction in range of movement of articulators, signs of apraxia of speech, or difficulty with coordination of the speech musculature were observed at that time, speech errors were classified as phonological paraphasia (word retrieval error) and not motor speech errors. Difficulties with BDAE repetition also were present, to the point where she resembled a person with conduction aphasia, e.g., Methodist Episcopal > *methodist es-pic, espis capih, espics …*; The barn swallow captured a plump worm > *the barn, the barn …* [after 2nd repetition by examiner]: *The barn … we have it* (i.e., they have barn swallows) … *the barn—swallow … something else, plump, plum*. [after 3rd repetition by examiner]: *the barn … swallow caught a—plum—worm.*

Phrase length, grammatical form, and auditory comprehension were relatively spared in both languages. Baseline scores on the BDAE (Goodglass et al., 2000) were similar in English and Shanghainese (Figure 2). Results from an

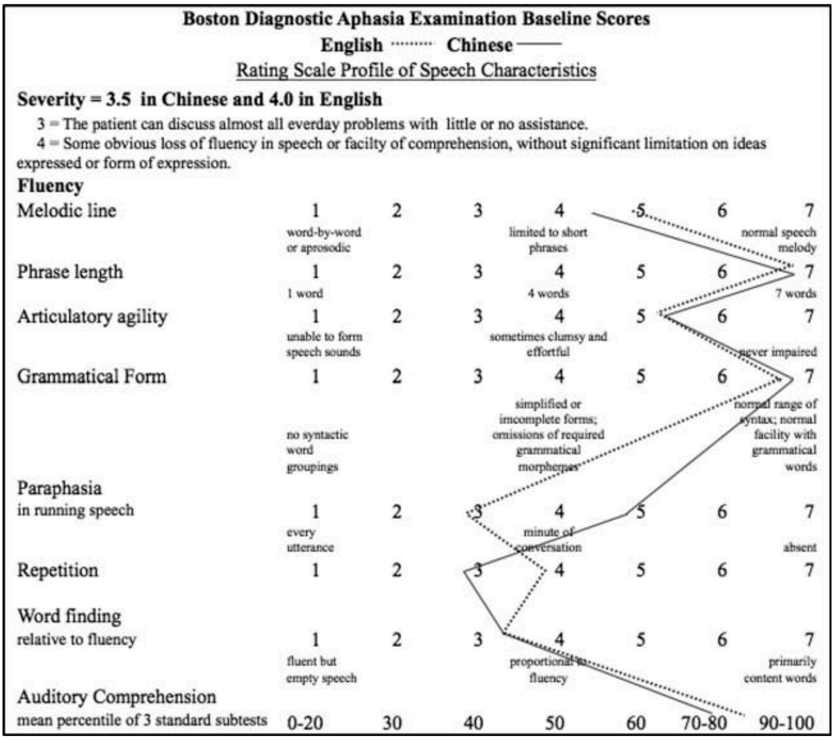

Figure 2. BYR's Boston Diagnostic Aphasia Examination (BDAE) scores (from Filley et al., 2006: © Taylor & Francis Group, 2006. Reprinted with permission).

electroencephalogram and magnetic resonance imaging (MRI) were normal for her age; however, single photon emission computed tomography (SPECT) showed mild hypometabolism in the left temporal and parietal regions. Subsequent to initial testing by our team BYR was diagnosed as presenting with PPA-LV (Filley et al., 2006; Hilger et al., 2011; Menn, Kong, Hilger, Ramsberger, & Yan, 2011).

Control speakers. Four elderly (65–95 years old), white, female speakers of English without aphasia or other speech or language impairments and with at least 12 years of education were recruited to provide typical language samples. One of these four women speaks English as a second language and has a slight German accent. The goal was to present raters with examples of what *good* Cookie Theft descriptions sound like from people with elderly, female and accented voices, so that the participant's slight accent (British English) and age would have less of an effect on the goodness ratings, which were the dependent measure. One Cookie Theft picture description was obtained from each of these four elderly typical speakers, and a fifth sample produced by a female with nonfluent aphasia was downloaded from AphasiaBank (MacWhinney, Fromm, Forbes, & Holland, 2011). The samples produced by the person with aphasia and from the participant with the German accent were used only during rater training. The remaining three samples were used in the rating procedure.

Raters. Raters were 15 female graduate students (estimated age range of 23–32 years) preparing for careers in speech-language pathology who were native speakers of American English without marked regional accents. Raters had no previous experience with the participant or any of the typical speakers and were not familiar with the purpose of the study.

Procedures

Picture description elicitations and transcription from the participant. A total of seven descriptions of the BDAE Cookie Theft (Goodglass et al., 2000) picture were digitally recorded (audio and visual) over the 27-month period BYR was followed by our team. With the Cookie Theft picture present in front of her, she was instructed to "tell me what is happening in this picture." No cues or leading questions were provided.

Spoken words were transcribed using Standard English spelling plus IPA phoneme symbols whenever standard orthography might have been ambiguous. Transcription conventions were similar to Ash et al. (2010) (see Appendix A). For analysis purposes, spoken Cookie Theft descriptions began after the instructions were given, when the participant began describing the picture. Descriptions ended when she indicated verbally or with head nod and eye gaze (towards the examiner) that she had completed the task. Comments on how she perceived her performances and verbalisations by the examiner were excluded from analysis.

Picture description elicitations from typical speakers. One Cookie Theft picture description was obtained from each of the four elderly typical speakers using the same instructions as described for the participant with PPA. The audio recordings of the typical speakers were made on a Mac OS X version 10.6.8 with Praat version 5.2.46. A fifth sample produced by a female with nonfluent aphasia was downloaded from AphasiaBank (MacWhinney et al., 2011) to approximate the hesitant but lexically adequate output of the participant with PPA. The samples produced by the person with aphasia and from the participant with the German accent were used only during rater training. The samples from the three native speakers of American English were used in the rating procedure; the sample from AphasiaBank and the sample from the speaker with the mild German accent were used only in the training procedure.

Measures

Analysis of picture descriptions: augmenting existing tools with a measure for speech sound errors. In a previous study of BYR (Hilger et al., 2011), we established that the very audible longitudinal decline in the quality of her Cookie Theft descriptions (which also distressed her greatly) was not adequately captured by existing tools for the analysis of connected speech such as the BDAE, the Linguistic Communication Measure (LCM; Menn, Ramsberger, & Helm-Estabrooks, 1994), or the Linguistic Communication Measure-Revised Cantonese (LCM-RC; Kong & Law, 2002, 2004). While these measures captured her lexical, syntactic, pragmatic, and morphological abilities, they did not respond to her increasing speech sound difficulties (phonetic and phonological errors and stutter-like onsets). The present study was motivated by the need to create and validate a measure that would track the progression of BYR's language decline. This additional measure was adapted from the Shewan Spontaneous Language Analysis (SSLA; Shewan, 1988), which does address the problem of such speech sound difficulties. The resulting battery of measurements, called the Linguistic Communication Measure-Speech Sounds

(LCM-SS), uses measures from the LCM (Menn et al., 1994), the LCM-RC (Kong & Law, 2002, 2004) to track total words, total content units (CUs), correct grammatical morphemes in CUs, semantic errors, and minutes of speech, plus the new measures of speech sound errors and words with repetitions, adapted from Shewan. Shewan's criteria for subdividing the speech sound production impairments as "phonetic" or "phonological", however, proved essentially impossible to apply reliably to BYR's output; therefore, we continue to refer to these errors collectively as "speech sound errors". We selected/adapted these components because they appeared to be appropriate to capture the most prominent characteristics of BYR's speech. See Appendix B for detailed descriptions of the coding procedures.

Intra-coder reliability for LCM-SS. The first author performed all of basic counts and computed all indices for each of the PPA-LV participant's Cookie Theft Descriptions. More than 1 month later, the same researcher performed all basic counts across all Cookie Theft Descriptions for a second time and computed all indices from those counts. Intra-coder reliability was determined by comparing the index scores from the first results with the second results using *Krippendorff's alpha* (Hayes & Krippendorff, 2007).

Cookie theft goodness rating. Audio files of the participant's seven discourse samples were extracted from the digital video recordings. These audio files, plus those of the three typical speakers and the two training speakers, were then uploaded into a program, run in MATLAB version R2010b, designed by the third author for rating of language samples.

Raters were first instructed in the use of the MATLAB program with the two audio files gathered for training purposes. They saw a screen containing the Cookie Theft picture, a *Play* button, a visual analogue scale with *Bad* labelled above the left end of the scale and *Good* labelled above the right end, and a *Next* button. Raters dragged a slider to move a bar along the rating scale to rate the goodness of the speech sample. Raters were simply instructed to listen to each recording and provide their impression of its *goodness*. No definition of *goodness* was provided to avoid introducing biases and hopefully to obtain a more ecologically valid rating of how speakers would be perceived by people with some training in speech-language-hearing science. Raters could not use the *Next* button until the speech sample finished. Raters listened to audio clips with over-the-ear headphones and were able to adjust the loudness to a comfortable level. Once the raters had demonstrated skill with use of the MATLAB program and rating scale (e.g., they used buttons and slider smoothly and they rated the speaker with aphasia as poorer than the elderly typical speaker with the British accent), they immediately began the rating task.

For the rating task, audio clips of each of the participant's seven recordings were presented three times each, along with the three audio clips from the three typical speakers (presented once each). These 24 audio clips were presented in a different randomised order to each rater. Thus, each of the 15 raters provided 24 ratings (three for each of the seven recordings of the participant, plus one for each of the three typical speaker recordings). Raters could briefly pause the program between ratings if needed, but all ratings were completed within a single 45- to 50-min session. Ratings were converted from the visual analogue scale to a number from zero to one.

Planned analyses. There were two planed analyses. First, LCM-SS counts and indices were plotted to reflect change over time and individual trend lines were created. Data for each measure were then correlated with corresponding trend lines and those with *R*-values of 0.50, or greater, were considered indicative of a reliable trends. Slopes were used to determine the direction of change, if any. Correlational and multiple regression analyses were then used to determine which LCM-SS counts/indices best accounted for the variance in *goodness* ratings.

RESULTS

Analyses of picture description

Transcriptions of BYR's Cookie Theft descriptions are provided in Appendix C. Basic counts and indices were calculated for the seven Cookie Theft descriptions produced by the participant with PPA (results are shown in Table 1). Scores were normalised so that they were on a comparable scale relative to each other. Normalised scores were plotted and trend lines were created for each measure. Visual inspection of the slope of the trend lines in Figure 3 for three of these six measures stood markedly apart from the others indicating a greater change over time: a 425% increase in sound errors (slope = 0.94; R^2 = 0.75); a 253% increase in the index of sound errors, which includes words with repetitions (slope = 0.07; R^2 = 0.89); and a 192% increase in words with repetitions (slope = 0.09; R^2 = 0.71).

Reliable, but smaller changes were evident for minutes of speech (65% increase; slope = 0.02; R^2 = 0.71), index of communicative efficiency (46% decrease; slope = −0.03; R^2 = 0.68), and correct grammatical morphemes in CUs (24% decrease; slope = –0.02; R^2 = 0.55).

There was no reliable trend for measures of total words or total CUs produced. Nor did any other grammatical (morphosyntactic errors, index of grammatical support, index of grammatical errors) or semantic measures (sematic errors, index of lexical effort, index of semantic errors) show a clear trend for change over time.

Intra-rater reliability of LCM-SS. High intra-rater reliability (k = 0.97) was determined verifying the consistency of LCM-SS coding of Cookie Theft descriptions.

Goodness ratings

The mean goodness ratings for the Cookie Theft descriptions produced by the three typical speakers were 0.95 (SD = 0.11), 0.95 (SD = 0.07), and 0.95 (SD = 0.06). In marked contrast, ratings for the participant's descriptions were about four standard deviations below that of the typical elderly speakers and declined from a mean high of 0.58 (SD = 0.18) to a mean low of 0.12 (SD = 0.09) (see Figure 4).

Intra-rater reliability of goodness ratings. Intra-rater reliability was determined to verify how consistently each rater scored the participant's Cookie Theft descriptions. κ-values ranged from 0.58 to 0.92 indicating that at least some raters were highly reliable and others were not as reliable. Outliers may have the effect of exaggerating the range of k value variability. To determine if this was the case, the mean k value (0.76) and SD (±0.11) were found. Ten of 15 reliability scores fell within one SD of the mean. An additional three scores were above this range and two scores were below it. Two SDs above and below the mean have values of 0.97 and 0.55; all reliability scores were within two SDs from the mean. These results indicate that no rater was a singular outlier in terms of rating reliability, so the results most likely reflect the variability of perception and rating.

TABLE 1
Linguistic Communication Measure—Speech Sounds (LCM-SS) counts and indices over time

Measure	*Time 1 + 0 months*	*Time 1 + 2 months*	*Time 1 + 7 months*	*Time 1 + 11 months*	*Time 1 + 15 months*	*Time 1 + 20 months*	*Time 1 + 27 months*
Basic counts:Total words (TW)	93	94	80	86	85	78	94
Total content units (CUs)	18	22	17	21	19	18	16
Correct grammatical morphemes in CUs (GM)	29	37	30	28	28	27	22
Morphosyntactic errors (ME)	5	1	2	6	1	5	6
Semantic errors (SemE)	3	2	0	2	3	2	3
Sound errors (SouE)	4	3	2	11	8	12	21
Words with repetitions (Rep)	12	11	14	13	24	19	35
Minutes of speech (Min)	1.17	1.47	1.47	1.42	1.68	1.5	1.93
Indices							
Index of lexical efficiency (ILE)	5.2	4.3	4.7	4.1	4.5	4.3	5.9
Index of communicative efficiency (ICE)	15.4	15.0	11.6	14.8	11.3	12.0	8.3
Index of grammatical support (IGS)	1.6	1.68	1.76	1.33	1.47	1.50	1.38
Index of grammatical errors (IGramE)	0.05	0.01	0.03	0.07	0.01	0.06	0.06
Index of semantic errors (ISemE)	0.03	0.02	0.00	0.02	0.04	0.03	0.03
Index of sound errors (ISouE)	0.17	0.15	0.20	0.28	0.38	0.40	0.60

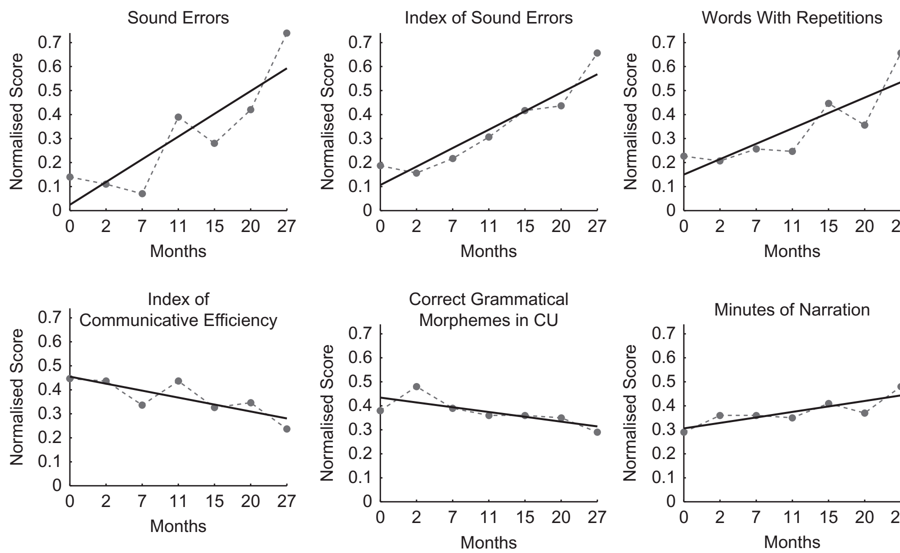

Figure 3. Six measures with reliable trends over time.

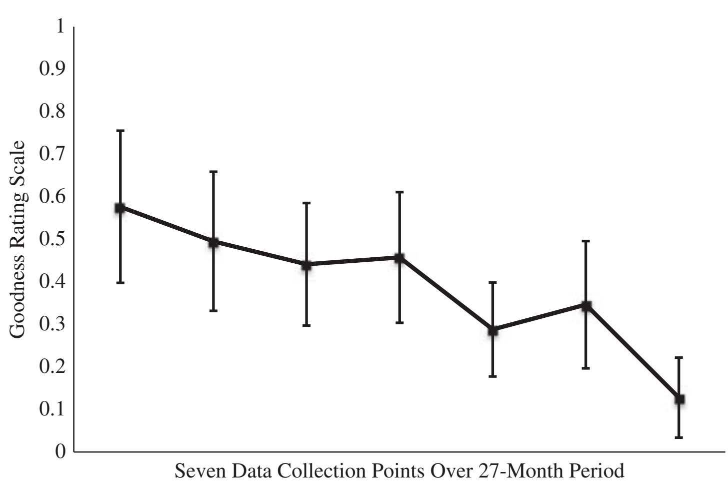

Figure 4. Mean goodness ratings and standard deviations over time.

LCM-SS relationship to goodness ratings

Individual raters used different amounts of the entire goodness rating scale continuum. To correct for this inherent variability, the goodness ratings were normalised such that the sum of squared values was equal to one for each rater. The result of this normalisation was that the probability density functions (PDFs) for each rater's scores were compressed and shifted around a common centre of mass, while maintaining information about the relative distances between ratings. To find an overall rating for each Cookie Theft description, the normalised scores were subjected to a multidimensional scaling analysis (MDS) (Abdi, 2007). Results of the MDS revealed

TABLE 2
Pearson correlations between LCM-SS indices and goodness ratings

LCM-SS index	*R*	*p-Value*
Index of sound error	−0.95*	.00096
Index of communication efficiency	0.93*	.00209
Index of grammatical support	0.54	.20731
Index of lexical efficiency	−0.46	.29675
Index of semantic error	−0.3	.51112
Index of grammatical error	−0.16	.73486

*$p < .05$; LCM-SS = Linguistic Communication Measure-Speech Sounds.

TABLE 3
Summary of regression analysis results for LCM-SS indices

LCM-SS index	*β*	*SEM(β)*	*p-Value*	*Power***
Intercept	0.5818	0.0227	.0000*	1
Grammatical error (IGramE)	0.2139	0.0535	.0162*	0.8427
Sound error (ISouE)	−0.8715	0.0613	.0001*	1

*$p < .05$; **$\alpha = 0.05$; Model: $y = 0.5818 + 0.2139 \times \text{IGramE} - 0.8715 \times \text{ISouE}$.

that the first principal component (PC1) accounted for 84% of the variance in the goodness ratings. Therefore, PC1 was projected to the normalised score space to reflect a scaled rating for each description. Next, correlational and multiple regression analyses were run to determine which of the indices best accounted for the variance in the overall scaled goodness ratings. For all statistical tests an alpha of $p = .05$ was considered the critical value for rejecting the null hypothesis.

Pearson's correlations showed that only two of the LCM-SS indices were highly correlated with the PC1 scores: index of communication efficiency ($r = 0.93$, $p < .002$) and the index of sound errors ($r = –0.95$, $p < .0001$) (Table 2).

Hierarchical multiple regression analysis with forward switching revealed that 99% of the variance in goodness ratings was accounted for by the LCM-SS index of grammatical error ($R^2 = 0.99$, $p < .0001$) and the index of sound errors ($R^2 = 0.98$, $p < .0001$) (Table 3).

DISCUSSION

The central objectives of this study were twofold: to create a measure that would capture the highly audible and debilitating changes in oral Cookie Theft descriptions in a participant with PPA-LV and evaluate the face validity of that measure—that is, its relationship to trained listeners' ratings of goodness for this participant.

Results from LCM-SS analysis are interesting in terms of both the features that worsened over time and those that showed small or no consistent pattern of change. Large increases over time in the number of speech sound errors and stutter-like sound repetitions were the most prominent negative feature of this participant's spoken output. While there was a small decrease in the use of free and bound grammatical morphemes, morphosyntactic components of the Cookie Theft descriptions cannot be characterised as impoverished; even in the later data grammatical errors remained low to almost absent. While the number of words spoken and the number of ideas expressed fluctuated from recording to recording, the efficiency of expression gradually decreased, as it took longer to complete Cookie Theft descriptions. No lexical errors were evident at any time although the frequency of word finding failure increased over time. This pattern of spared and impaired language skills is consistent with previously reported as key features of the PPA-LV (Gorno-Tempini et al., 2011; Rohrer et al., 2010; Wilson et al., 2010). Rohrer et al. (2010) suggested that people with PPA-LV have more extensive involvement of posterior elements of the language network that results in a core deficit affecting manipulation of phonological information. They go on to say that longitudinal investigations of PPA-LV might be able to provide empirical data to test this proposed pattern of disease evolution.

The PPA-LV case we have described here is the first reported case showing that a speech sound production deficit remained the primary impairment to the language network over the course of the disease. If this striking pattern is found in other cases, it may warrant further subdivision of PPA-LV into cases with and without heavy sound production difficulties. Future longitudinal investigations combining sensitive behavioural measures, such as the LCM-SS, and imaging analyses should further elucidate the distinct linguistic features of this disease and its anatomical substrates as well as contribute to our understanding of the relationship between components of the language network.

The LCM-SS adds sensitive measures of speech sound aspects of the language network to the syntactic and semantic measures of the LCM and LCM-RC, but it does not require the discrimination between phonetic and phonological difficulties. This may contribute to reliability of coding since this distinction is often difficult to make. The LCM-SS also simplifies the way stuttering-like speech is quantified. Instead of counting each repeated phoneme, which would be a task almost impossible to do online during a clinical evaluation, words with repetitions are counted. This method of accounting for stuttering-like speech was also shown to be reliable and it contributed to the index of sound errors' significant correlation with perception of quality. The speech sound measures of the LCM-SS may have value in the analysis of connected speech samples produced by people with other forms of PPA or other speech sound disorders, but this needs to be demonstrated.

Analysis of spoken picture description may be an important means of quantifying features of spoken output both when diagnosing and when tracking changes in people suspected of having PPA. Additionally, it is essential that the method utilised is sensitive to phonological/phonetic errors and speech repetitions, as well as lexical and morphosyntactic features, since these are key features in the three variants of PPA.

While the intra-rater reliability of the LCM-SS was established in reporting the results of this case study, inter-rater reliability was not established. Since the LCM-SS was developed using the methods of three measures (i.e., LCM, LCM-RC, and SSLA) with high inter-rater reliability (i.e., 0.956, 0.926, and 0.977) we predict that the inter-rater reliability of the LCM-SS will be comparable; however, it will be important to demonstrate this before the LCM-SS is used for comparisons across participants and raters. Furthermore, since the LCM-SS is ultimately intended to have clinical applicability, the time necessary for clinicians to develop reliability should be determined and compared to the training time required for other connected speech analysis methods.

The second purpose of this study was to determine whether the results of this analysis of picture descriptions were related to raters' perceptions of discourse quality. Listeners' ratings indicated that the quality of this participant's Cookie Theft descriptions continually declined over the course of the 27 months that she was followed. LCM-SS indices proved to be highly correlated with and predictive of qualitative ratings. Because BYR's morphology, lexicon, and syntax remained nearly stable, we infer that what these listeners were responding to was the decline in sound production abilities that is captured by the new indices of speech sound errors and repetitions.

An intuitive goal for analysis of connected speech is that the results are grounded in what is clinically meaningful. To be clinically meaningful, results must be more than just significant, they must reflect perceived changes. The procedures in this study accomplish this by evaluating the analysis of picture descriptions based on what listeners perceive as changes in the goodness. Future investigations are encouraged to utilise this or a similar method to demonstrate construct validity of analysis methods.

As is always the case, results of this longitudinal case study must be interpreted cautiously. Detailed transcription and coding instructions for the LCM-SS are provided in Appendices A and B in the hope that others will explore its value in evaluation of people with PPA-VA, but it also must also be tested on people with other forms of PPA that primarily impact semantics and morphosyntax, and with other types of connected speech samples.

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APPENDIX A

LCM-SS transcription conventions

A comma represents a pause following an intonation contour that would be transcribed by a comma in ordinary orthography.
A dash at the end of a partial or full word indicates an abrupt stop to the final sound.
A dash between partial or full words indicates stuttering-like repetitions of phonemes, phoneme clusters, or words.
A colon after a letter indicates a sound that the speaker produced for a longer time than sounds normal in typical speech, as judged by the researcher.
Within an utterance, a short silence is indicated by one dot, a medium length of silence is indicated by two dots, and a long silence is indicated by three dots.
Anomalous pronunciations are given in phonological notation set off by slashes: / /. If the word she intended to say is clear given the context, the word appears orthographically after the phonological production in parentheses and quotations: ("word").
Notes, comments, and glosses of anomalously pronounced words are given in parentheses: ( ).
Overlapping speech lines by two speakers are indicated by greater than/less than signs around the words that overlap: < >.

APPENDIX B

Detailed instructions for implementing the LCM-SS

Counts

Identifying total number of words

(1) Count all of the words in the transcription.
(2) Include: incorrect words, neologisms, phonological paraphasia, semantic paraphasia, full word repetitions, self-corrections (however, false starts on a word that is eventually produced are not included), irrelevant statements, digressions, habitual statements, comments, and fragments that seem to be identifiable as broken-off words.
(3) Exclude: Cross out all hesitation noises, interjections, untranscribable mumbles, false starts on a word that is eventually produced, and a subject's direct responses to questions or probes from the examiner. (When the subject produces a schwa vowel sound, it can be either the hesitation *uh* or the indefinite article *a*. When *a* would be grammatically correct, give the subject credit for it and count it as a word.

Identification of content units (CUs)

(1) Underline all of the correct informative words. *Correct* means that the word would contribute to the listener's understanding of the scene being described and would not give a listener an incorrect mental picture of the scene. *Informative* means that the word adds significantly to the information that a listener is obtaining for building a mental picture about the scene. Informative words may be any part of speech.
   (i) The following are not informative words: neologisms, semantic paraphasia, and phonological paraphasia that result in real nontarget English

words (i.e., "burger" for "burglar"). A word that echoes a word supplied by the examiner is not considered an informative word.

(ii) The following should be included as informative words: broken-off words that a listener would be able to reconstruct correctly with the help of the context a subject provides in the description.

(iii) Personal pronouns used correctly are counted as informative the first time they appear. Subsequent uses are treated like other repeated words (see step 3).

(2) Put parentheses around each informative word and the phrase of words surrounding the informative word. The description should be divided into the phrases or natural word groups surrounding each informative word. If there is more than one informative word in a set of parentheses, make shorter phrases such that there is one informative word in each set of parentheses.

(3) The parentheses are the boundaries of potential CUs.

(4) In a single picture description (i.e., the "Cookie Theft"), count only one instance of each informative word, if some are used more than once. Choose the CU to keep based the richness of the surrounding phrase; keep the informative word and its CU that has the most surrounding words and morphology, and cross out the informative word with the less rich CU. In a language sample describing a sequence of actions, count repeated words as informative when their re-use is necessary to make clear who or what is being referred to. Also, if a word is re-used and refers to a different person or object from the earlier referent, it should be counted as an additional correct informative word because it is adding to the listener's understanding.

(5) If a CU contains an incorrect or incomplete word that has not been fully self-corrected, cross it out. If a CU contains a self-correction sequence, cross out all of the sequence except for the subject's best effort.

(6) All CUs now contain correct informative words and the "supporting words" that make up the rest of the CU phrases.

Identification of bound and free grammatical morphemes within CUs

(1) Put a box around (by hand) or italicise (on a computer) the correct *bound* grammatical morphemes. Word "endings" are considered bound grammatical morphemes (i.e., plural, third person—*s*, past tense—*ed*, possessive—*'s*, progressive—*ing*, etc.). Contracted grammatical elements are also considered bound grammatical morphemes (i.e., negative—*n't*, contracted "verb + is" forms—*'s* as in "he's going" and—*'re* as in "we're going", future tense "will + verb" form—*'ll* as in "I'll go", etc.). Additional bound grammatical morphemes include prefixes such as *un*- such as "unaware" and "undoing".

(2) Exclude and put a line through: semantically incorrect endings (e.g., plural ending when singular item is being referred to, use of verb + −*ing* when the action referred to is completed. In a syntactically ill-formed phrase like "you have going", exclude either the "have" or the –*ing*, as they cannot both be correct.

(3) Put a box around (by hand) or italicise (on a computer) the correct *free* grammatical morphemes. These are also known as closed-class words and consist of articles, pronouns, conjunctions, prepositions, interjections, and numbers. Additional closed-class words are the "is/are" part of a progressive verb as in "is going", "is/are" part of a future tense verb as in "are going to go", modal

verbs (i.e., “would”, “could”, “should”), negation words “not” and “no” as in “not aware” and “no one”, and the verb infinitive form marker “to” as in “to eat”.

(4) Count all bound and free grammatical morphemes within CUs that have been circled/italicised.

Identification of morphosyntactic errors

(1) Look for morphosyntactic errors in the complete language sample as opposed to inside CU boundaries.
(2) Highlight in one colour all errors of morphology (i.e., incorrect bound grammatical morphemes, also referred to as word endings and prefixes).
(3) Highlight in the same colour all syntactic errors, including incorrect forms of words (e.g., “you have going” which should be “you have *gone”* or “you *are* going”) and words that are missing (e.g., “I gave it you” which should be “I gave it *to* you”). When a phrase is abandoned and a second phrase is started that is syntactically incongruous with the first, do not count it as a syntactic error (e. g., the phrase “and the water was the sink is over” has no syntactic errors although it does have an abandoned word “overflowing”, which is categorised as a sound error).
(4) Include as errors changes in tense and aspect within a phrasal unit, however do not count as errors changes in tense and aspect across the sample as a whole. It is difficult to determine if the latter changes are correct or incorrect pragmatically during a picture description, therefore determination of correct and consistent use of tense and aspect is kept at a phrasal level.
(5) Syntax also includes word order. Highlight each word group that was not spoken in the correct, grammatical order. For instance, the phrase “she was going pool to laps swim” contains three syntactic errors: “going pool” is missing the preposition “to” and either article “the” or “a” depending on context, and “laps” should come after “swim”. The correct form is: “she was going *to the/a* pool to *swim laps*”.
(6) Count all highlighted morphosyntactic errors.

Identification of semantic errors

(1) Look for semantic errors in the complete sample as opposed to inside CU boundaries.
(2) Highlight in a second colour all semantic errors. These will often be considered semantic paraphasia (e.g., “spoon” for “fork”). The wrong gender terms are used in the example “the sister is, she is uh climb up on the stool”, spoken in reference to the Cookie Theft picture. Count this as a single semantic error because the gender error (i.e., “sister” for “brother” and “she” for “he”) is repeated and not corrected.
(3) Include first attempts at a word that is eventually produced correctly such as the phrase “she, he was …”, produced in reference to a boy.
(4) Include attempts at a word that is not eventually produced correctly, even if the speaker recognises the attempts are incorrect (e.g., “Skip? No. Trip? No. Anyway …” in reference to a boy falling off a stool).
(5) Count all of the highlighted semantic errors.

Identification of sound errors

(1) Look for sound errors in the complete sample as opposed to inside CU boundaries.
(2) Highlight in a third colour all sound errors. Sound errors are phonological paraphasia, neologisms, jargon segments, words that are broken off (e.g., "cook jar" for "cookie jar"), and words produced incorrectly that are eventually corrected (e.g., "getting the goo-coo-cookie" has one sound error, "goo" and also has repetition of "cookie").
(3) Count all of the highlighted sound errors.

Identification of repetitions

(1) Look for repetitions in the complete sample as opposed to inside CU boundaries.
(2) Circle or highlight all full and partial word and phrasal repetitions.
    (i) For full word repetition, count each instance of the repeated word.
    (ii) For partial word repetition, count each word that has a partial word repetition(s) as opposed to counting the number of repetitions. Partial repetition may be produced quickly, therefore counting the number of repetitions may be difficult. If a nonword is repeated, count this as both a sound and a repetition error.
    (iii) In the example, "water is-is-is is-is o-over the-the-the fu-fu-floor", two words have full word repetition ("is" and "the") and one word has a partial word repetition ("over"). The partial word repetitions of "floor" are considered sound errors and would be counted as two instances of sound errors during the identification of sound errors.
    (iv) Count each phrasal repetition as a single unit (e.g., "the sister is, she is" contains one phrasal repetition, even though the exact words were not used the second time). Do not count repeated phrases that provide additional information during the second production (e.g., the repetition in "and the girl, his sister wants" provides clarification as to the girl's identity and should not be treated as a repetition error).
(3) Do not include repetitions when the subject repeats to correct a semantic error in the first utterance (e.g., "to get hi- get her one"), as these are captured during identification of semantic errors.
(4) Do not include repetitions of words that have sound errors and are eventually produced correctly (e.g., "/sku/uh stool"). These are captured during identification of sound errors. In the sequence "s-sə-sith-thi-think sə-something-thing", the first "think" part is counted as both a sound error (because of "sith") and a repetition error (because of "thi-think"). The second, "something", part is counted only as a repetition error because there are no incorrect phonemes in this production.
(5) Count all of the partial words with repetitions and full word and phrasal repetitions.

Indices

Index of lexical efficiency (ILE): calculated the same way as LCM; total number of words divided by the number of words in content units.

Index of communication efficiency (ICE): calculated the same way as LCM-RC; number words within content unit divided by the number of minutes for total language sample.

Index of grammatical support (IGS): calculated the same way as LCM-RC; the number of correct closed class words and affixes divided by the number of words in content units.

Index of grammatical errors (IGramE): total morphosyntactic errors within CUs divided by number of CUs; indicates the level of morphosyntactic impairment during narration.

Index of semantic errors (ISemE): total semantic (verbal) paraphasias within CUs divided by the number of CUs; indicates the level of semantic impairment during narration.

Index of sound errors (ISouE): sum the total sound errors and the number of words with repetitions, then divide by the number of CUs; indicates the level of sound production impairment during narration.

APPENDIX C

Transcriptions of BYR's cookie theft descriptions

Time 1 + 0 mos.

GR: Tell me everything you see going on in that picture.BYR: Yeah.

*[begin analyses]

BYR: The boy is uh, trying to get uh, to the cookie cookie jar. And the sister is, she/iz/('is') uh.. climb up on the/sku/-uh-stool. And the sister wants one of the/ka?/-uh cookie too and ask of the, the boy to uh get hi- get her one. And uh, uh. Meanwhile the, the mother is uh doing dishes an-an ('and-and') she's she's uh daydreaming and uh water-water uh- on the fl-fl-floor and uh an ('and') it's a nice day outside. The window is o- is uh open and so must be nice uh outside.. and she is/noʊ/('not') uh a-aware at all about uh what the ki-kids are doing.

*[end analyses]

GR: That's fine.

Time 1 + 2 mos.

LM: so we're going to ask you again to tell us what you see (puts Cookie Theft picture in front of BYR) in this picture.

*[begin analyses]

BYR: um the mother i:s doing, uh di – washing the, uh. dishes and uh she- is thinking and uh and she- uh she uh i-i-is not aware the, water is uh. f-flooded the floor and the wo-BYR:it is a nice day,.. the win-win-window is open, an' children were. uh trying to, um get some – cookies uh in the. um... the higher… (gesture, something horizontal, laughs) the.. on a upper. shelf

LM: mhm!

BYR: and they, the boy- boy had to, uh:. uh step on a – sto-ol to, reach, to the… n – the cookies, and u:m, and sh- he is going to fall uh – fall down, and the uh, the, the sister was saying I wanted one- I want, one too…

*[end analyses]

LM: that’s fine. OK, thanks!

Time 1 + 7 mos.

LM: So, again, if you would look at that picture and tell us the story of what’s happening. Okay?
BYR: Mhm…BYR: You want me to-to-to < Yeah. Okay>.
LM: <you-you can start. Yeah. >
*[begin analyses]
BYR: um. the mother is uh doing the, um.. dishes and she’s thinking and then she, th-uh the uh water was uh.. uh.. um… (hand movement, laughs) uh.. uh… uh, the water. is um… the faucet, she-she let the, f-fauceth. um.. (laughs) the sink, f-full and it was spilling: down to the: floor. And the children were. um, boy and a girl and the uh, the boy is uh.. uh… i:s getting the co-cookies a-and then he’s gonna/duv/fa- fall from the stool, and um. and the girl, his sister, wa-wants to uh.. to uh- ask him to-to get, get her a cookie
*[end analyses]
BYR: God!
LM: Good!

Time 1 + 11 mos.

LM: So, can you tell us the story in English, <of > what’s <here?>
BYR: <Yes,> <yes. >
*[begin analyses]
BYR: Um, mother is-is um. uh. washin. dishes. and she is thinking and uh and she have/eɪd/(possibly ‘made’?) the water.. flooded. And the children were-were s-.. um.. try to reach to the/kʊk taɪt/cookie.. cookie (points to husband, laughs) jar, and the girl, the girl said brother give me/ʌnə/(‘another’) one, and he/I-/the/I-/the the stool is-is gonna um: fall down. The um… mm.. (makes hand gesture) skip? No. Trip? No. (laughs) Anyway, huh-/I/-/I/-/I/he is going to fall down.
LM: mhm
BYR: The w-w-win (‘window’)/I/-/I/-it’s a duh- warm day the- one win-window is ob^ (‘open’)
LM: mhm
BYR: They have all sh-short sleeves… and cups.. all done. (appears to be comment on state of the cups, not a comment about finishing the description)
*[end analyses]
LM: Mhm. That’s fine.

Time 1 + 15 mos.

LM: Alright, so what’s going on here?
BYR: (clears throat) xxx (mumbled sounds) okay,
*[begin analyses]
BYR: Aaahm… the g- the, the, mother is: doing.. doing.. w-washing dish-ishes, and she is uh.. is uh… dreaming.
LM: (nods, but BYR continues to look at the picture and does not see this)
BYR: and the the the w-w-w-wa-water was- was… (~8 sec pause)
BYR: /ði/(‘the’)/ðə/(‘the’) sink i:s. over-… (5 sec pause)

BYR: She should-d-d <turn-turned off the-the… (~5 sec pause; small gesture while looking at picture, might be writing)>
LM: <(nods, not seen) >
BYR: the/waər/(‘water’)
LM: mhm.
BYR: And the w-. w-w-. w-w-. window is o-o-. is o-over uh, open, and the boy is-is getting-getting cookies from the. she-shelf, and-and the- buh- uh-. his sister wants a-a/k^ki/(‘cookie’) also, An’ then she- he was-as going to f-fall. fall-all down, because the the stool is-is.. is uh um... is going to fall down.
*[end analyses]
LM: (nods) Right.. Okay.
BYR: (laughs and looks up)

Time 1 + 20 mos.

GR: (Opens stimulus notebook) Okay. So, you’ve seen this <picture>
BYR: <Yes > uh ch- (laughs)
GR: Many times.
BYR: Yeah.
GR: What I want’cha t’do is tell <me >
BYR: <Yeah.>
GR: what’s happening in the picture.
*[begin analyses]
BYR: /ɪ-ɪ/-it’s a nə-nice. day. warm./thæ-/the w-wi-winda is open.
GR: (nods, but BYR isn’t looking – she’s continuing to look at the picture)
BYR: and uh. uh. s- the mother is-is. uh.. thy bishin. w-washing dish-dishes
GR: mhm
BYR: and uh- s- an’ she is s-sə-sith-thi-think sə-something-thing because of the water-r/ɔ/-/ɔ/-/ɔ/(‘all’?) um. fl-flow over the to the uh the. uh, uh./fwɔ/- <floors> an’ she didn’t even see it
GR: <mhm>
GR: mhm
BYR: and the childrens went uh… t-tr-ch-. try to get th:e cookies
GR: mhm
BYR: and the- the. the um the the/bo/(‘boy’)- is going to fall down from the stool
GR: mhm
BYR: and uh. the sister want to say heluh- also. uh- also/wəwə/- duh.. uh/k^p-kok-ko-//kʊ-/cookies (screws up face, nods while saying ‘/kʊ-/cookies’)
*[end analyses]
GR: mhm! mhm! Good, good.

Time 1 + 27 mos.

BYR: (breathing audibly throughout Cookie Theft description)
GR: And I want you to look at it and tell me everything that’s going on.
BYR: xxx um. brea-breathe you know?
GR: Yeah. Do you want to wait until you get your breath?
BYR: (breathes audibly and coughs, waits ~15 s)
*[begin analyses]
BYR: mama is-is pa-pa.. sh- wa- washin. d-.. dis-is,

GR: mhm.
BYR: an-u-tus.. water the-the… is-is st- fili-. s:-sli-. uh. the-the.. uh.. the windows is o-. open. and it's ni-. days. ousi-si-si outside,..
GR: (nods, BYR does not see, BYR looking at the picture)
BYR: the children have, a boy and-and a. gir- (brings hands together, then apart again to resting)
GR: (nods, BYR does not see, BYR looking at the picture)
BYR: ther-there's um girls and uh.. the guh-, the boy is a-a a cook-cook jar-ar-ar, all-ga-a a-a again-s-s-… o-a-a the-the-the. lid and-and he-he is uh-get he is… getting the goo-coo-cookie uh to-to the. sister,..
GR: mhm.
BYR: and uh the- he i-is-is. go. go-going to.. fall-lo-lo ('fall off'?) from the uh stoo-,
GR: mhm.
BYR: and the hallo-of wofe,. uh.. wa- water is-is-is is-is uh. o-over the-the-the fu-fu-floor
*[end analyses]
GR: Mhm.
BYR: (exhales deeply)
GR: Alright. Good.