

# Fluency in primary progressive aphasia - logopenic variant

## A fluência na afasia progressiva primária logopênica

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### ABSTRACT

This research is a case report which aims to perform a longitudinal analysis of fluency of a subject with Primary Progressive Aphasia (PPA), Logopenic variant. The method of analysis was based on seven speech therapy sessions of a 61-year-old patient diagnosed with PPA. The data was analyzed qualitatively in light of Enunciative-Discursive Neurolinguistics. The results showed that the patient has the logopenic variant of PPA, with the following characteristics: preservation of speech grammar structure, preserved comprehension of single words and phrases during conversation in dialogical interaction. Speech flow difficulties were marked by occurrences of repetitions of longer and more unusual words, phonological paraphasias, non-fluent pauses and anomies. The analysis also pointed to a progression of symptoms, with increasing number of repetitions of different forms of occurrence. These modifications suggest an inversely proportional relationship between oral fluency and disease progression in which speech flow tends to decay. This scenario is relevant for subjects' language because it influences their social interaction, i.e., the less fluent their speech, the more affected they are as speakers.

**Keywords:** Aphasia; Primary progressive aphasia-logopenic; Language; Neurolinguistic; Neuropsychology

### RESUMO

Esta pesquisa é um estudo de caso que tem como objetivo analisar, longitudinalmente, a fluência de um sujeito com Afasia Progressiva Primária (APP) Logopênica. O método de análise baseou-se em sete sessões de atendimento fonoaudiológico de uma paciente com 61 anos de idade, diagnosticada com APP. Os dados foram analisados de forma qualitativa, a partir da Neurolinguística Enunciativo-Discursiva. Os resultados sugerem que a paciente apresentava a variante Logopênica da APP, com as seguintes características: fala com estrutura gramatical reservada, manutenção da compreensão de palavras isoladas e de frases preservadas, durante a conversação na interação dialógica. As dificuldades de fluência referiram-se às ocorrências de repetição em palavras longas e menos cotidianas, parafasias fonológicas, pausas disfluentes e anomia. A análise também apontou modificação progressiva nos sintomas, com aumento do número de repetições e alteração de sua forma de ocorrência. Essas modificações parecem indicar uma relação inversamente proporcional entre fluência de fala e avanço da doença, em que a fluência tende à deterioração. Esse cenário assume aspecto relevante na linguagem do sujeito na medida em que influencia na sua interação e papel social, ou seja, quanto menos fluente é o discurso, mais essa condição afeta sua posição de falante.

**Descritores:** Afasia; Afasia progressiva primária logopênica; Linguagem; Neurolinguística; Neuropsicologia

Research conducted in the Undergraduate Program in Speech Therapy, Universidade Federal de Santa Catarina – UFSC – Florianópolis (SC), Brazil, based on a final term paper by one of the authors, entitled Primary Progressive Aphasia: a case study.

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**Conflict of interests:** No

**Authors' contribution:** *KPS*: main author; study conception and design, literature review, collection, analysis and interpretation of data, drafting and revision of manuscript; *DCR*: coauthor, study conception and design, literature review, data collection and analysis; *APS* advisor, research design, timetable, collection, analysis and interpretation of data, literature review, drafting and revision of manuscript, final revision.

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**Received on:** 3/12/2015; **Accepted on:** 8/17/2015

## INTRODUCTION

Primary Progressive Aphasia (PPA) is characterized by progressive deterioration of language through gradual atrophy, which can remain isolated from language for up to ten years<sup>(1)</sup>. PPA is considered part of a group of rare neurodegenerative diseases, including frontotemporal dementia that appears in individuals aged between 45 and 70. Although Progressive Aphasia subtypes were described more than 100 years ago, in the cases studied by Pick, Sériex, Dejerine, Franceschi, and Rosenfeld<sup>(1,2)</sup>, it was not until the 1980s that more systematic studies started being conducted on the subject.

In 1982, neurologist Mesulan was one of the pioneers that researched the phenomenon. He described six early cases of PPA<sup>(3)</sup>, and highlighted the initial presence of language change in the clinical picture, with absence of global cognitive impairment signals (i.e., dementia). The modern description of PPA<sup>(4)</sup> as a clinical entity, apart from degenerative dementias, aroused debate in the literature about its legitimacy as a distinct syndrome. Thus, because of its progressive nature and similarities with other dementia diseases at an early stage, there is a recurrence of the use of the term “dementia process” in its definition<sup>(4)</sup>.

There is no consensus in the literature regarding the classification of PPA syndromes or subtypes<sup>(5)</sup>. However, there are four major variants: Agrammatic Primary Progressive Aphasia (PPA-A), Semantic Primary Progressive Aphasia (PPA-S), Mixed Primary Progressive Aphasia (PPA-M) and Logopenic Primary Progressive Aphasia (PPA-L). PPA-L, the subject of the present study, is associated with damage in the first few convolutions of the left temporal lobe and, therefore, among the aforementioned variants, it is the most frequently associated with Alzheimer’s Disease (AD), because they both have similar symptoms.

Inaccurate distinction between PPA and AD leads to up to 15% of errors in diagnosis as a result of low frequency of cases and aetiological ambiguity between the two diseases<sup>(4)</sup>. This is due to the fact that the clinical pictures of both PPA and Alzheimer’s Disease are difficult to define at their early stages<sup>(6)</sup>. However, the diseases can be distinguished when neural aspects and endophenotypes between AD and PPA-L are compared<sup>(7)</sup>.

Speech fluency is usually identified as one of the facilitating factors for differentiation between PPA and dementias when the former is investigated<sup>(6)</sup>. In PPA-L, both grammar and comprehension remain relatively preserved. However, the speech of subjects with PPA-L is marked by constant hesitation, long anomic pauses, pathological repetition of sentences<sup>(4)</sup> and difficulties in phonological choices<sup>(2)</sup>. Phonological difficulties, i.e., the effort assigned to lexical access and the inability for sentence repetition, appear to be the result of damage to the left posterior temporal gyrus, or more specifically, a thinning of this area, which is characteristic of the syndrome<sup>(7)</sup>.

It should be noted that speech flow is understood, in such cases, as the result of production of words per minute. In this

sense, the pursuit of speed and lexical access were relevant for classifying PPA variants; PPA-A is also referred to as Non-Fluent PPA (NFPPA) and PPA-S, as Fluent PPA (FPPA). In other words, vocabulary production per minutes (understood as speech flow) is consistently low in the logopenic and grammatic subtypes and high or excessive in the semantic subtype, with object naming as possibly the most significant difficulty in neuropsychological testing<sup>(4)</sup>. Although the logopenic subtype is not characterized as FPPA, it can be seen that speech flow, considered from a discourse perspective, is one of the relevant factors to the diagnosis of the variant, since the speech of patients with PPA-L is marked by hesitations and pauses that interrupt “the flow of conversation and give their speech a nonfluent quality”<sup>(7)</sup>. Longitudinal studies<sup>(8)</sup>, especially on the logopenic variant of PPA, can shed light on disease progression.

Speech flow, therefore, is an important aspect of language for the characterization and diagnosis of PPA, and it has been understood quantitatively, with a focus on production. There are no studies analyzing fluency in PPA from a dialogical perspective and with a qualitative approach. Therefore, this study, conducted from the perspective of enunciative-discursive neurolinguistics<sup>(9)</sup>, can bring theoretical contributions to the field, as this theory favors the analysis of the speech of subjects in meaningful contexts of production, as well as their work on language. Thus, the aim of this study was to make a longitudinal analysis of speech flow of a subject with PPA-L.

## CASE REPORT

This study deals with the qualitative and longitudinal analysis of Roberta’s case (not her real name), a retired 61-year-old accounting clerk, who completed vocational secondary education. Roberta was initially diagnosed with Alzheimer’s Disease (AD) in 2011, and she was diagnosed with PPA only in March 2012. The patient complains of difficulty in articulating speech and trouble in finding words.

The results of clinical MRI (03/03/2011), EEG (06/07/2011) and SPECT (26/09/2011), indicated, respectively: (1) “small image with low signal intensity on T2, juxtacortical lesion in the middle frontal gyrus on the right, related to calcification or chronic hemosiderin deposit”; (2) “irregularities and disorganization of the base rate, with mixed pattern of frequencies and wide variability in the amplitude modulated by theta activity, reactive to eye opening and closing. Hyperventilation caused nonspecific, slow bilateral response”; (3) “severe hypoperfusion in the anterior projection of the left temporal lobe and moderate hypoperfusion in the middle third and posterior projections, ipsilaterally; mild hypoperfusion in the projection of the left occipital cortex.”

Transversal data collection was performed between March 2012 and March 2014 and took place at the University Hospital (UH) of a federal university in Brazil. The episodes reported in this study were recorded in clinical speech therapy sessions,

conducted from the perspective of Enunciative-Discursive Neurolinguistics. The data, which comprise four episodes of reading and three episodes of spontaneous speech, were transcribed and analyzed qualitatively. However, they show frequency of occurrence of events. Data were transcribed by following these conventions<sup>(10)</sup>: (( )) for insertion of researcher's comments; + for marking every 0.5 pause; ( ) to inform the length of pauses longer than 1.5s; ---- for syllabication; : for marking vowel stretching and " for marking interrogation.

This study was approved by the Research Ethics Committee of the *Universidade Federal de Santa Catarina* (UFSC), technical report no. 21084913.6.0000.012. Both the patient and her guardian signed an informed consent form.

## Reading

### Data 1: Episode 1R (4/5/2012)

**Context:** Roberta read aloud a magazine headline (*Isto é* magazine).

#### Turn Interl. Utterance

- |   |    |  |
|---|----|--|
| 1 | R  | swimming English <b>horse-man-ship</b> tennis soccer is becoming more common to find children who <b>barely le (+) barely left</b> school and already <b>fol follow</b> a mini executive agenda with appointments that continue throughout the day there are some words that I can't pronounce   |
| 2 | IV | that you can't pronounce "which are they" let's note them down   |
| 3 | R  | ((Roberta resumes reading)) <b>fol (+) follow</b> executive <b>mi-ni</b> agenda with appointments that continue throughout the day parents' intention is to subject their children to these routines is to make them highly prepared adults for the competitive <b>modern (+) modern</b> world (...) parents' intention is to subject their children to these routines is to make them <b>highly (+) highly (+) prepared</b> for the competitive world modern world the price to pay for such a big effort however can be high <b>still small children (+) still small these children</b> start showing grown-ups' problems stress it is a swap that does not <b>pay (+) off</b> claims the <b>psycho psycho-thera (+) therapist</b> |
| 4 | IV | the biggest names are harder"  |
| 5 | R  | They are   |

### Data 2: Episode 2R (9/20/2012) (5 months later)

**Context:** Reading aloud a news story about wine.

#### Turn Interl. Utterance

- |   |   |  |
|---|---|--|
| 1 | R | ((reading)) scientists from the University of Texas in the United States noted that this <b>an-ti-o-xi-dant en encourages</b> the body to <b>re release</b> a very welcome hormone <b>a-di-po-ne-c-tin</b> |
|---|---|--|

produced by **fa (+) fat** cells it favors the action of insulin stopping gain weight gain and the chance of success of type 2 diabetes is a great discovery but the challenge now is to establish the optimal dose of **res-ve-ra-trol** for consumption and increase its use by the body since it is easily eliminated **says chemist says chemist** André Souto from the **pon-ti-fi-cal** catholic university of Rio Grande do Sul so the recommendation remains two glasses of wine a day for men and one for women interesting right? "

### Data 3: Episode 3L (11/13/2013) – (1 year and 7 months later)

**Context:** Reading aloud the story "The Princess and the Frog."

#### Turn Interl. Utterance

- |   |   |   |
|---|---|---|
| 1 | R | once upon a time in a land far away there was a beautiful princess, independent and full of <b>self self-esteem she came across (+) she came across</b> a frog while she was <b>contem (+) contemplating</b> the <b>na nature</b> and thought of what a what a <b>wonderful (+) wonderful</b> lake by her castle <b>was was</b> relaxing and environment-friendly (+) then the frog jumped into her lap and said beautiful princess I've been a very handsome prince an evil witch cast a spell on me and I became this disgusting frog a kiss from you however can turn me back into a <b>ha hand handsome prince (5.0)</b> turn me back into a handsome prince and we can get married and <b>build (+) build</b> happy home in your beautiful castle your mother could come live with us and you could <b>pre prepare</b> my dinner wash <b>my (+) wash my</b> clothes raise our children and we would be happy ever after that night as she tasted frog legs <b>(3.0)</b> tasted frog legs together with a creamy onion sauce <b>and some (+) and some</b> fine white wine, the princess smiled thinking to herself "no way, not over my dead body" ((laughter)) |
|---|---|---|

### Data 4: Episode 4L (3/15/2014) (2 years later)

**Context:** Reading informational text "The water in the world."

#### Turn Interl. Utterance

- |   |   |  |
|---|---|--|
| 1 | R | water in the world water is very important for our life it is present in many <b>acti acti acti activities</b> of our daily life in our daily hygiene when we bathe wash hands before meals we <b>bra bru (+) bru brush</b> our teeth etc it is also present in our leisure when we refresh in the river on the beaches or in the pools water is <b>al also fun-da-men-tal</b> for hydration of our body <b>when we drink (+) when (+) when we drink</b> |
|---|---|--|

water and other **liqui liqui liquids** we also use **on on on domestic (+) domestic** chores such as washing clothes cleaning floors etc. there is a great deal **ab about** water it is present in the **mi (3.0) minor** movements of our body like winking after all we are **com-pos-posed** basically of water in more than 70% of our body water is an essential element in our lives but drinking water is not **avail available (+) end (+) endless end-less-ly** it is a limited **recourse resource** water is also threatened by pollution by contamination and **cli cli climate** changes that human beings **are are** causing bringing great danger to the health and **well-be (+) well-being** of man so each of us **shou should** use water more **eco economically**.

### Spontaneous speech

#### Data 5: Episode 1S (4/5/2012)

**Context:** Talk about the headline she read, referring to the stress that children suffer from overload of activities.

#### Turn Interl. Utterance

- |    |    |   |
|----|----|---|
| 1  | IV | what do children do all day? “What does Ana [granddaughter] do?”  |
| 2  | R  | goes to school right “(...) I don’t know if there are children with lots of activities I don’t know children like that  |
| 3  | IV | what activities can children can do besides classes”  |
|    | R  | Ana comes home from school and takes off her uniform and goes cycling until later in the evening  |
| 6  | IV | does she go alone or do you keep watching her”  |
| 7  | R  | No, I watch her (+) when I don’t the neighbor across the street keeps an eye on her... no, she can go alone, right” but my street is a quiet, few cars drive by, most often in the late afternoon |
| 8  | IV | do you help her with homework”  |
| 9  | R  | her mother helps more often because sometimes I try but there are days when I can’t   |
| 10 | IV | but do you help or have you stopped now”  |
| 11 | R  | I help.   |

#### Data 6: Episode 2S (11/13/2013) (1 year and 7 months later)

**Context:** Spontaneous conversation with the therapist, after a period of absence in the clinic. R tells how she feels at the moment.

#### Turn Interl. Utterance

- |   |    |   |
|---|----|---|
| 1 | R  | <b>it’s it’s it’s</b> like <b>(3.0)</b> sort of <b>(3.0)</b> for speaking it’s (+) there are days when I speak well but there are days when (+) it’s very difficult |
| 2 | IV | Are you having speech therapy there”  |

- |    |     |   |
|----|-----|---|
| 3  | R   | no <b>no I’m not talking (2.0) I’m not having it</b>  |
| 4  | IV2 | Have you discontinued”  |
| 5  | R   | <b>I ha have</b>  |
| 6  | IV2 | oh I thought you were still having the therapy  |
| 7  | R   | No  |
| 8  | IV2 | Are you seeing a psychologist?  |
| 9  | R   | I see a psychologist, speech therapist and <b>(3.0)</b> what is it called” <b>(4.0)</b> I forget the name of it |
| 10 | IV2 | you used to have painting classes, are you still having them?   |
| 11 | R   | Yes, I am   |
| 12 | IV2 | you were seeing a psychologist, a neuropsychologist, who worked on your memory                                  |
| 13 | R   | then she sent me to another (+) to another <b>(3.0)</b> person”   |
| 14 | IV2 | the one I’m seeing <b>is :::</b> what’s it called “ <b>(3.0)</b> occupational therapist”                        |
| 15 | R   | Right   |
| 16 | IV2 | And have you been reading? Writing.. how’s that?”   |
| 17 | R   | Right   |
| 18 | IV2 | And have you been reading? Writing.. how’s that?”   |
| 19 | R   | <b>To writhe (+) write (+) it’s it’s</b> harder but I always do it  |

#### Data 7: Episode 2S (3/15/2014) (2 years later)

**Context:** Spontaneous conversation.

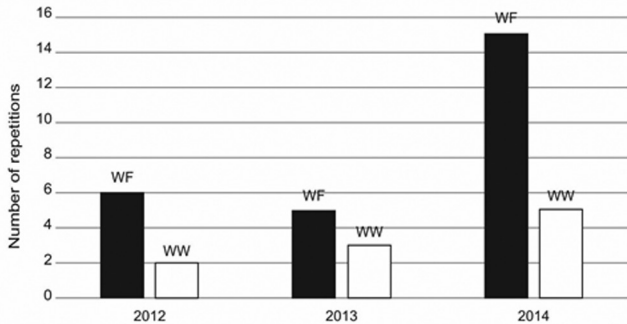
#### Turn Interl. Utterance

- |    |    |  |
|----|----|--|
| 1  | IV | How are you?   |
| 2  | R  | <b>well:::</b> I’m better on some days (+) I’m just OK on some days  |
| 3  | IV | Uh-huh   |
| 4  | R  | but it’s OK it’s OK  |
| 5  | IV | Are you still seeing the speech therapist?   |
| 6  | R  | see the therapist (+) see <b>the::: theratis theratisp</b>   |
| 7  | IV | Uh-huh   |
| 8  | R  | and there’s <b>pilakes</b>   |
| 9  | IV | oh OK, I thought you had stopped seeing the therapist.   |
| 10 | R  | no no (...) there’s a machine <b>that::: (5.0)</b> can’t say what machine it is (+)and::: <b>(6.0)</b> there’s a machine that gives shock shocks |
| 11 | IV | hmmm you said so last time and it’s good”  |
| 12 | R  | yes uh-huh I think it’s fine (+) much better with her  |
| 13 | IV | and the difficulty in speaking”  |
| 14 | R  | <b>well:::</b> there are days I can <b>spes speak</b> well, other days are::: hard   |
| 15 | IV | gets stuck”  |
| 16 | R  | stuck <b>uh::: (4.0) uh::: (5.0) uh::: (3.0)</b> ((shakes her head to deny)) gets stuck and <b>no:::</b>   |
| 17 | IV | and it’s hard?”  |
| 18 | R  | uh-huh it’s hard (..) <b>I swif swee switch</b> the words  |

## DISCUSSION

By analyzing the four episodes of the reading task, it could be seen that Roberta’s reading was marked by repetitions and syllabifications, which, while not predictable as to their occurrence, were somewhat systematic. By analyzing the repetitions in more detail, it is clear that they are frequent disfluencies in reading and they had the following characteristics<sup>(11)</sup>: a) Word fragments such as “fol follow” (episode 1R T1), “re release”, “fa (+) fat” (episode 2R , T8), “contem (+) contemplating”, “na nature”, “hand handsome” (episode 3R, T2), “al also”, “bout about” “mi (3.0) minor” (episode 4R T2), among others; b) Whole words such as “modern(+)  
modern” (episode 1R, T1), “wonderful (+) wonderful”, “was was”, “build (+) build” (episode 3R, T2), “are are “(episode 4R, T2), among others; c) Utterances like “says chemist says chemist” (episode 2R , T8), “she came across (+) she came across,” “wash my (+) wash my” (episode 3R , T2), “well-be (+) well-being” (episode 4R, T2), among others.

The repetition word fragments and whole words were the most frequent, with increasing occurrence during the course of the syndrome (Figure 1). These two types of repetition are directly proportional to the length of PPA, that is, the more advanced the syndrome, the higher the occurrence of this disfluency.



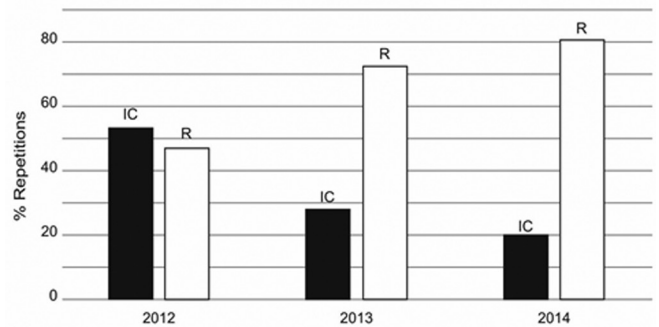
Note: WF = word fragment; WW = whole word

Figure 1. Type of repetition in word

Roberta’s reading rate remained unchanged during data collection, but her reading rate was perceived as slower in episodes 3 and 4 than in previous episodes. There were several pauses in all episodes.

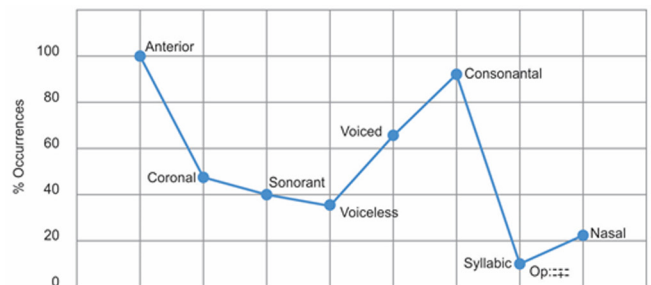
It was found also that while dysfluency in complex and/or irregular words or utterances with complex and/or irregular words accounted for more than 50% of disfluency occurrences in 2012, this figure did not amount to even 20% in 2014 (Figure 2).

As for phonological aspects, it was observed that the repetitions occurred before anterior phonemes (+anterior), especially in +consonant, voiced phonemes (Figure 3). In this sense, Roberta tended to have greater difficulty in producing phonemes with these features. Therefore, as can be seen later



Note: IC = irregular and complex; R = regular

Figure 2. Type of repeated word



Note: Op. = Opening

Figure 3. Phonological context of repetition

in the spontaneous speech tasks, occurrences of paraphasias also tended to occur before that phonological context.

Roberta had intelligible speech in spontaneous speech tasks, which is indicative that her oral language was still preserved. She accurately reported her granddaughter’s activities and showed very fluent speech in episode 5. In episodes 6 and 7, unlike episode 5, Roberta showed difficulties in maintaining speech flow. For this reason, she made longer pauses and repetitions, so that she could produce her utterance. Except for the repetition of word fragments, such as in “ha have” in episode 6, T5, Roberta’s disfluencies, including pauses, do not seem to be related to a specific type of word, but they proved to be more related to an enunciative functionality, as language strategies. The literature has categorized these aspects as prospective and retrospective repairs, which show speakers’ effort in the development of what will be said<sup>(12)</sup>: prospective repairs for failed memory (anomie), with pauses, as in “(3.0) :: she bought (+) bought a machine to (+) shock shocks (+); retrospective repair for correction of paraphasias, as in “no, I’m not talking (2.0) I’m not having it”; “swif swee switch the words”. It was noted that in order to maintain fluency, Roberta relied on the utterance of her interlocutor so that she could develop her own utterance (episode 7S, turns 17-18): (IV: and it’s hard? R: uh-huh it’s difficult); she sometimes resorts to her interlocutor.

Another observation from a qualitative point of view, which seems to point to a worsened clinical picture, are changes in repetitions. Unlike in 2012 and 2013, in 2014 Roberta started to repeat the same word fragment more often, whole word and



part of the utterance, for example, “acti acti acti activities”, “bra bru (+) bru brush”, “liqui liqui liquids”, “clim clim climate”, “on on on” and “when we drink (+) when (+) when we drink” (episode 4R, T1). It was observed that disfluency occurred both in regular words and in complex and irregular words. However, the occurrence of disfluency in utterances with complex and/or irregular words were most significant in the first episodes, accounting for more than 50% of disfluency instances.

Qualitative analysis of Roberta’s reading skills and spontaneous and dialogical speech revealed characteristics of PPA-L at the early stages of the disease<sup>(4)</sup>. In addition to the lesion found by MRI, which matches the location described in the literature - damage to the angular gyrus and the first left temporal convolutions<sup>(13)</sup> - Roberta’s speech showed preserved grammatical structure, preserved comprehension of individual words and preserved phrases, during conversation in dialogic interaction. Moreover, she had more difficulty in producing long words, both in oral language<sup>(7)</sup> and in reading; she chose to read syllable by syllable instead of the whole word. Likewise, less commonly used words appeared as repetitions and reading difficulty<sup>(7)</sup>.

The above data showed that as Roberta’s disease progressed for two years after diagnosis, her dysfluency increased, and tended to increasingly occur at regular words, showing a progressive degeneration of her language system, as far as reading is concerned. However, Roberta’s speech cannot be analyzed only in terms of deficit. It should also be mentioned that while the repetitions and syllabication showed deterioration of language, they also revealed Roberta’s efforts on language. In this regard, the concept of language used by Enunciative-Discursive Neurolinguistics<sup>(9)</sup> should be emphasized: language is seen as work shared among interlocutors so that they can construct and interpret meaning. Roberta performed epilinguistic work on language<sup>(14)</sup> by hesitating, rephrasing, negotiating “articulatory gestures” for reading and demonstrating, thus, the subject’s relationship with language itself. Roberta showed to be aware of her difficulties, acknowledging her inability to pronounce a few words. The increase in these difficulties, however, did not prevent Roberta from understanding the text, but placed her in the role of apprentice, although an apprentice who “works” on the language, according to her own words: “*it seems that I am learning to speak*”<sup>(15)</sup>. Thus, it can be seen that Roberta’s disfluencies in speech also showed the enunciative-discursive strategic function and demonstrated, in fact, her epilinguistic work on language. Through disfluencies, Roberta maintained her enunciation flow, making herself understood by her interlocutor<sup>(11)</sup>.

## FINAL COMMENTS

The two-year longitudinal analysis of Logopenic Progressive Aphasia showed qualitative and quantitative changes in symptoms, with increased number of repetitions (in speaking, writing

and reading) and changes in how it occurred (in reading). These changes seem to suggest an inverse relationship between speech fluency and disease progression, whereby fluency tends to worsen. This scenario becomes relevant in subjects’ language, as it influences their interaction and their social role, because the less fluent speech is, the more this condition affects their role as speakers.

Another point that seems to draw more attention in language deficits is the set of aspects related to speech disfluency. Such disfluency is expressed through repetitions, anomies, long and frequent pauses and difficulty in lexical and phonological access (paraphasias and phonological exchange). Seen from this angle, speech disfluency expresses more than low production of words within a time slot; it discloses several difficulties and linguistic symptoms which leads the subject to disfluent speech. At the same time, they reveal strategies and conditions for continued production of speech, even if more slowly, because they expand the time of linguistic elaboration and re-elaboration. In this sense, this research indicates that understanding disfluency in subjects with LPPA can be crucial to better understand language in use, as well as its evaluation and therapeutic process.

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