Case Report

Relato de Caso

Language in Frontotemporal Dementia: an analysis in light of Enunciative-Discursive Neurolinguistics

A linguagem na Demência Frontotemporal: uma análise à luz da Neurolinguística Enunciativo-Discursiva

Daniel de Martino Ucedo[1]
Karoline Pimentel dos Santos[1]
Ana Paula de Oliveira Santana[1]

Keywords

Speech, Language and Hearing Sciences
Neuropsychology
Frontotemporal Dementia
Language
Pathology

ABSTRACT

The aim of this case study was to perform a cross-sectional analysis of spontaneous speech of a patient with Frontotemporal Dementia (FTD). For this purpose, four speech and language therapy episodes, from 2012 to 2014, were selected, transcribed and analyzed in light of Enunciative-Discursive Neurolinguistics. The analysis showed, as the patient's FTD status progressed, that he used different semiotic strategies, e.g., use of repetition and gesture during speech production. It also highlighted the importance of the interlocutor's role of prompting the patient to express verbal meaning. Thus, it can be concluded that the recognition of the strategies used by the patient in favor of his role as a speaker, during interactions, is what enables and legitimates his role.

Descritores

Fonoaudiologia
Neuropsicologia
Demência Frontotemporal
Linguagem
Patologia

RESUMO

Este estudo de caso objetiva analisar longitudinalmente a fala espontânea de um indivíduo com Demência Frontotemporal (DFT). Para isto, foram transcritos e analisados quatro episódios de terapia fonoaudiológica de um indivíduo com DFT entre 2012 e 2014 à luz da Neurolinguística Enunciativo-Discursiva. A análise evidenciou, ao longo da progressão da DFT, as diferentes estratégias semióticas utilizadas pelo indivíduo, como o uso discursivo da repetição e do gesto, bem como o lugar de importância do interlocutor para a promoção do fazer-dizer do indivíduo. Nesse sentido, conclui-se que o reconhecimento, na interlocução, das estratégias utilizadas pelo indivíduo em favor de sua posição de falante é o que viabiliza e legitima esta posição.

Endereço para correspondência:
Karoline Pimentel dos Santos
R. Aracuã, 140, Pantanal, Trindade,
Florianópolis (SC), Brazil,
CEP: 88040-310.
E-mail: karol.pimentel@gmail.com

Received: October 08, 2016

Accepted: April 10, 2017

Study carried out at Universidade Federal de Santa de Catarina – UFCS, Florianópolis (SC), Brazil.
[1]Universidade Federal de Santa de Catarina – UFSC, Florianópolis (SC), Brazil.

Financial support: nothing to declare.
Conflict of interest: nothing to declare.

INTRODUCTION

Frontotemporal dementia (FTD) is the fourth most frequent dementia worldwide[1] and the second most common in individuals under the age of 65[2]. FTD is characterized by changes in personality, behavior and language, as a result of the strong presence of atrophy in the frontal and temporal lobes at the early stage of the disease[1-5]. Although FTD can be clearly identified in topographic distribution analyses, individuals with FTD present very diverse neuropsychiatric changes, depending on location of lesions, degree of brain atrophy in the affected region and medical history[6,7]. In this scenario, language processing is one of the functions most frequently affected by FTD[1,2]. This occurs because language, either written or spoken, offers evidence about the cognitive status of an individual[8], even though the correlation between speech and cognition is not a direct one[9].

Individuals with FTD may present imbalance in their daily activities, resulting not only from a behavioral change, but also from the manifestation of "disorganized" speech as well as loss of motivation for communication or excessive speech production[10]. In this respect, according to the neuropsychological literature, individuals with FTD have, above all, difficulty in using language in social settings. Their speech breaks conversational and turn-taking rules and fails to take previous utterances into account[10]. Over time, there is a gradual decrease in verbal fluency, presence of circumlocutions, repetitions (palilalia, echolalia, amimia) and stereotypy, as well as a decline in verbal comprehension, until the final stage of the disease, in which there is total absence of communication[2].

In the speech-language clinic, based on the historical and social perspective of language, the dialog held between the individual with FTD and his therapist - about the daily interactions of this individual in other social contexts - can offer further knowledge about the dynamics of interlocutor roles in the construction of meaning[11]. This occurs because the therapist considers FTD patients to be in charge of their utterances and encourage them to perform and report everyday tasks that they deem as meaningful. Above all, their aim is to foster conversational strategies that support verbal interaction, because such strategies are inherent in this type of interaction.

Within this perspective, Enunciative-Discursive Neurolinguistics (EDN) considers the social environment, even if outside the body, as a crucial element of an individual's cognitive dynamics[11]. Thus, language in seen in EDN as forming individuals and their various social roles, hence it primarily seeks to reposition individuals in a place where they can perform discursive management of their own speech. Also, EDN takes into account the multimodality of speech and language and, therefore, the analysis of language includes both oral and written speech and non-verbal contributions offered by an individual during conversation[11]. Consequently, individuals have broader possibilities of signification and linguistic reorganization while dialog is taking place.

Few studies to date have addressed language in FTD from a dialogical perspective[8]. Conversely, the vast majority describes language changes without, however, looking into the context where these changes occurred[6]. Therefore, while considering the social nature of language, the objective of this study is to perform an analysis - from the perspective of EDN - of the oral language of an individual with FTD. As a method, a two-year, cross-sectional case study was performed between 2012 and 2014. For the present paper, four episodes from this period were selected and analyzed in light of EDN.

CLINICAL CASE PRESENTATION

The participant in this research is called Heitor (fake name). He was born in the southern region of Brazil in 1950. He is 64 years old and married. Heitor is an accounting technician and partner in a cafeteria. Before his illness started affecting his daily life, he worked as a sales representative in the cosmetics industry. He was initially diagnosed with suspected FTD or Alzheimer's disease (AD) in 2010; in 2011, he received the definitive diagnosis of Frontotemporal Dementia.

The results of the clinical examinations by Magnetic Resonance Imaging (MRI), held on 3/1/2011, and Single Photon Emission Computed Tomography (SPECT), held in 9/27/2010, indicated, respectively: "Decreased radiopharmaceutical concentration (hypoperfusion), mild degree, in the left frontal pole; moderate to strong hypoperfusion in the left prefrontal region; hypoperfusion, moderate degree, in the left temporal lobe" and "prominence of intracranial subarachnoid space mostly along the temporal convexity and the left frontal convexity, and the T2 weighted images show hyperintense signal in the subcortical white matter of the left temporal lobe and also of the frontal lobe on the same side, and thinner gyri of the cerebral convexity." Finally, "the images show hippocampus sections with normal morphology, appearance and signal intensity".

The neuropsychiatric evaluation showed the following results for the MMSE (Mini–Mental State Examination): 15/30 on 5/31/2012; 08/30 on 2/7/2013; 09/30 on 5/9/2013; 05/30 on 11/7/2013; 08/30 on 6/5/2014. The following neuropsychological descriptions also composed the individual's clinical picture: disorientation in time and space, poor speech, lack of initiative, excessive use of monosyllabic words (with echolalia and perseveration), expressive aphasia.

In the speech-language therapy, the initial complaints brought by his wife were: difficulty in recalling the name of objects, memory lapses (including names of people), reduced practice of reading and doing crosswords (habits he had had for years), in addition to decreased speech production. As reported by his wife, Heitor ran away from home a few times, was inattentive when crossing the street, and was uncommonly "stubborn". In the shower, he started confusing personal care products, e.g., he used hair conditioner to wash his body instead of a soap bar. During meals, he refused to use cutlery and ate very quickly, despite the high temperature of the food.

In the initial speech and language evaluation, carried out on (29/3/2012), Heitor showed reduced speech production and use of specularity to express himself and make himself understood. The concept of specularity[12] characterizes the dialogic relationship and occurs when one of the interlocutors "mirrors" the speech of the other. For example, in language acquisition, children introduce parts of adults' utterances into their own utterances.

Heitor was referred to weekly END-based speech and language therapy sessions, which lasted for 45 minutes each. The sessions had activities designed to stimulate oral and written speech production by means of diverse discursive genres (book summaries, maps, advertising flyers of supermarkets, lists, personal accounts, life history narratives, drawings, news, crossword puzzles, etc.).

For the present research, four video-recorded episodes from his language-speech therapy sessions were selected and analyzed. These episodes took place at the Speech-Language Pathology Clinic, Federal University of Santa Catarina, between August 2012 and August 2014. They were considered to be representative of changes to the patient's language. The episodes were transcribed[1] and analyzed from the perspective of Enunciative-Discursive Neurolinguistics[11]. This study was approved by the Research Ethic Committee (protocol no. 02674912.0.000.0121). The participant's legal guardian signed an Informed Consent Form and an Authorization Form for Use of Image/Video Recording.

The episodes below feature dialogs between therapist(s) and the patient that participated in the present research. The dialogic topics, described in detail in each episode, include issues relative to the participant's everyday life: activities and daily habits (episodes 1 and 3), favorite television shows (episode 2), recent news that have affected his routine (episode 4).

Episode 1 - 6/4/2012, one year after diagnosis of FTD.

Dialog between the interlocutor (INT, the therapist) and Heitor (H), developed around Heitor's personal account about his daily activities in a speech-language therapy session.

Turn	*Initials*	*Transcript*
1	INT	What have you done today? What time did you wake up? Do you remember? What time did you wake up?
2	H	Seven-thirty.
3	INT	What was the first thing you did?
4	H	I had some coffee.
5	INT	Do you remember what you had coffee with?
6	H	I ate a cookie.
7	INT	And what did you do after that?
8	H	I brushed my teeth.
9	INT	What else? Did you go out? Did you watch TV?
10	H	Watch TV.
11	INT	What was showing on TV? Don't you remember?
12	H	No.
13	INT	And what else did you do? Don't you remember? What about lunch? What did you have for lunch today?
14	H	I had... An egg... (makes a pause)
15	INT	An egg...
16	H	Two eggs.
17	INT	Oh, you like them...
18	H	I do! And rice.
19	INT	Rice.
20	H	And green beans.
21	INT	I like green beans. Don't you ever eat beans?
22	H	No.

[1] The transcript was aimed at preserving the typical characteristics of the language spoken by the participant, e.g. pauses and repetitions. The researcher included comments about the enunciative scene.

Episode 2 - 8/6/2012, two months after the first episode.

The therapist/researcher (INT) and Heitor (H), during a speech-language therapy session, talk about an episode of the Brazilian soap opera "Avenida Brasil", which had been broadcast at the night before the therapy session. In that episode, the character Carminha pretends she is jealous of her husband Tufão. She threatens to go away from home even though she actually does not mean to leave.

Turn	*Initials*	*Transcript*
1	INT	What about yesterday? Last night, did you watch it?
2	H	I did.
3	INT	Let's write down the names of the characters here... Carminha. And who else is in the soap opera? What's her husband's name?
4	H	Tufão.
5	INT	And what happened last night? Did Carminha want to stay with Tufão or separate from him?
6	H	Se... Tufão.
7	INT	What happened to Carminha last night? Did she want to stay home or go away?
8	H	No, she didn't... She didn't want to go away.
9	INT	No?
10	H	No.
11	INT	What did she want then?
12	H	Tufão.
13	INT	Did Tufão want to go away?
14	H	No.
15	INT	Didn't he, either? Did he want to stay with her?
16	H	Wanted to stay with her.
17	INT	Does she want Tufão to stay with her?
18	H	Stay with her.

Episode 3 - 5/6/2013, eleven months after the first episode.

Interlocutors - therapist (INT1) and therapist (INT2) - and Heitor have a dialog, during a speech-language therapy session, about Heitor's daily activities.

Turn	*Initials*	*Transcript*
1	INT1	What did you have for breakfast? This morning at breakfast...
2	H	A... What's it called? *((tries to make hand gestures but gives up and looks at INT2.))*
3	INT2	Bread? *((Browses the pages of a notebook with images of food))*
4	H	No, no... cheese. *((looks at notebook))*
5	INT2	Ham too?
6	H	Cheese and ham.
7	INT2	Sandwich?
8	H	Sandwich
9	INT2	Write cheese, ham, bread, then. ((*dictating))*
10	H	*((H writes the words cheese and bread on the sheet of paper))*
11	INT1	Ham too? *((after realizing H had not written the word ham))*
12	H	No.
13	INT2	What have you had for lunch today, sir?
14	H	Cheese. *((points to the written word "cheese"))*
15	INT2	Lunch...
16	H	No, no. *((makes a hand gesture for the word no))*

Turn	Initials	Transcript
17	INT2	Haven't you had lunch yet? *((makes a facial expression of surprise))*
18	H	Cheese. *((points to the written word "cheese"))*
19	INT2	You had cheese for breakfast. What about lunch? *((makes a facial expression of surprise))*
20	H	No.
21	INT2	Rice, beans...?
22	H	No, no. *((points to the written word "cheese"))*
23	INT2	Haven't you had lunch yet?
24	H	No! Cheese, OK.
25	INT2	Have you had cheese for lunch? *((makes a facial expression of surprise))*
26	H	No, no... Look. Cheese. *((points to the written word "cheese"))*
27	INT2	At noon...
28	H	Cheese... cheese... *((tries to make a hand gesture but gives it up))*
29	INT2	Bread too?
30	H	No.
31	INT1	No bread...
32	H	No.
33	INT1	Any orange juice?
34	INT2	Before you came over here... didn't you eat anything?
35	H	Didn't eat anything.

Episode 4 - 6/3/2014, two years after the first episode.

Context: Dialog during speech-language therapy, about the bus strike taking place at the time and the strike at the university.

Turn	Initials	Transcript
1	INT	Did you realize there was a strike last Wednesday?
2	H	Last Wednesday.
3	INT	Do you have a say on that? About the bus drivers' strike? Do you think it´s a good or a bad thing?
4	H	Here, look... *((long pause))*
5	INT	Don´t you ever take the bus?
6	H	*((reaches for his wallet and fetches his bus pass))*
7	INT	Is it the only thing you have to show the driver?
8	H	*Yes... ((after a long pause, he looks at INT))*
9	INT	But you can't go around town when there's a strike.
10	H	*((silence))*
11	INT	But what do you think about the strike? Don't you mind it?
12	H	Whatever... No, look, ... er... *((long pause, then points to the window))*
13	INT	At university?
14	H	No, here... At university, right? *((long pause, then points to the window; after that, catches the activity folder as if looking for something))*
15	INT	What happened here? If there's a bus drivers' strike, students won't be able to come. There was also a strike at the university.
16	H	Yes, strike at university *((points to the window))*
17	INT	Is the strike a bad thing?
18	H	Yes... *((long pause, then looks at INT))*
19	INT	Why do you think it's a bad thing?
20	H	It's... here... *((long pause, makes a hand gesture for 'I don't know', then looks at INT))*
21	INT	Nothing works?
22	H	Nothing works.

DISCUSSION

Episode 1 shows that Heitor's oral language is characterized by simplified utterances. It appears that he has difficulty in using the personal pronoun (I) and subject-verb agreement for first person, for example, in turns 9 and 10 (INT: *Did you watch TV?; H: Watch TV)*. This difficulty is "resolved" by mirroring the therapist's speech[12]. Difficulties with verb agreement were not systematic, as the patient was able to use subject-verb agreement at other times, without further difficulty, as can be seen in turn 6 (*H: I ate a cookie)*. Lack of enunciation and turn-taking also occur in the conversation (*int: What else? - turn 11)*. Traditionally, Heitor's speech can be characterized as having difficulty in lexical access; it is marked by pauses and dysfluencies, in addition to losses at the semantic-lexical levels[10] (*H: ate an egg....((pause)) rice, green beans*). It is deemed, therefore, as "reduced speech"[13]. In this way, Hector's account of what he ate had to be mediated by the interlocutor ("*you like them…"* - turn 17) so that information could be fully input into the dialog. The interlocutor referred to time progression in the utterance in order to articulate the information contained in Heitor's speech. Thus, turns 1, 5, 7, 13 and 21 ("*What have you done today?*", *"What time did you wake up?", "...what you had coffee with?", "And what did you do after that?", "And what else did you do?" Don't you remember? What about lunch? "What did you have for lunch today?", "I like green beans."* "*Don't you ever eat beans?*") helped Heitor produce utterances in turns 2, 6, 8, 14 and 22 ("*Seven-thirty*", "*I ate a cookie*", "*I brushed my teeth*", "*I had... An egg...*", "*no*"). Possibly, the intended utterance would be "*I woke up at seven-thirty, ate a cookie, then brushed my teeth. Later, I had an egg for lunch without beans, because I don't like beans."*

His difficulties are clearly indicative of disorders when he attempted to access functional and lexical terms which, ultimately, directly affect his level of pragmatic competence and make him more dependent on the interlocutor's utterances. These difficulties also reveal a "telegraphic talk" (also called reduced speech), which can also be seen here as an alternative strategy he used in order to cope with his difficulties in selection and combination of words[9]. FTD is a degenerative disease; therefore, one year after diagnosis of the disease, it can be seen that Heitor cannot produce fluent utterances without the interlocutors' help. In the first episode, he manages to achieve his intentions in discourse through by relying on pauses and on his interlocutor's collaborative interventions.

In Episode 2, notably, there are occurrences of "repetition". However, although these repetitions present verb agreement errors, they are not the mere transposition of the interlocutor's speech and, therefore, they do not represent a deficit per se, nor are acts of repetition, such as perseveration and echolalia, deemed as pathological. On the contrary, they indicate, in the midst of linguistic deficits resulting from FTD, the patient's speech attempts by selecting and using his interlocutor's speech with a view to producing his own utterances[14]. The analysis of the dialogs shows that, in Heitor's speech production, these repetitions occurred when the interlocutor's speech and Heitor's enunciative intention coincide, i.e., when the utterance to be

repeated is in agreement with what Heitor wishes to say, as can be seen in turns 15 and 16, 17 and 18 *("INT: Did he want to stay with her? H: Wanted to stay with her"; INT: Does she want Tufão to stay with her? H: Stay with her"*). When there was a difference between the propositions, i.e., between the utterance actually made by the interlocutor and Heitor's intended utterance, he answered negatively, as in turns 13 and 14 (*INT: "Did Tufão want to go away?", H: "No"*). It can therefore be said that Heitor's repetition can be considered as a dialogic strategy that is similar to the concept of specularity[12]. This is because, in order to actively produce speech, Heitor received the support of the utterance of his interlocutor to (re)formulate his own utterance, and he takes a speculative approach to his interlocutor's speech.

In Episode 3, approximately one year after episodes 1 and 2, one can see a significant dissolution of Heitor's language. There is an almost total absence of spontaneous speech and greater dependence on the interlocutor's speech. In this scenario, the joint effort of the interlocutors was essential for collaborative construction of signification, which accesses the construction of meaning. In the episode in question, the interlocutors attempt to express Heitor's intended utterance, e.g., in turns 3, 5, 21, 29 and 34 (*"Bread?", "Ham too?", "Rice, beans...?", "Bread too?", "Before you came over here? Didn't you eat anything?"*), by negotiating the construction of meanings.

What is noticeable is that, at this stage of the disease, the role that the interlocutor is required to play is even more active, by offering possible themes for construction of meaning. In this way, poor interpretation of linguistic mechanisms used by patients with FTD can lead to misunderstandings in conversation and, as a result, the absence of speech-related therapeutic actions required for them to complete their utterances. This is due to the fact that the interlocutor finishes the other person's utterance to fill a gap in their difficulty[15]. The concept of finishing involves the wholeness of values of aspects that are invisible to the other person.

Also in this dialog, it should be noted that the interlocutors often have to *finish* an utterance to construct meaning, for example, when Heitor makes use of the word *"cheese"* in turns 14, 18, 24, 26, 28: at first, the interlocutors did not understand that Heitor had not had lunch because they assumed given the time, that Heitor would have had lunch already. Therefore, INT2 asks the same question in several ways in utterances 13, 15, 17, 19, 21, 25, 27 and 34 *("What have you had for lunch today, sir?", "lunch...", "Haven't you had lunch yet?, "You had cheese for breakfast. What about lunch?", "Rice, beans...?", "Have you had cheese for lunch?", "At noon...", "Before you came over here...* didn't you eat anything?*"*). In response, Heitor can only speak the word *"cheese"* spontaneously, although he tried to modify the forms of enunciation by using negation in turn 24 (H: *No! Cheese, OK.*) and gestures, pointing to the written word to refer to the context of breakfast in turns 22 and 26 (*"No, no. ((points to the written word "cheese")), No, no... Look. Cheese. ((points to the written word "cheese"))*. Although the word "*cheese"* may, at first, be analyzed as perseveration, the analysis of the dialog shows that repetition of the word cheese is an important enunciative position assumed by Heitor. It is through this position that he confirms the only meal he had had as a mechanism to deny having had another meal, in this case the lunch he had been asked about. In this regard, it is the role of the therapist to observe what an individual speaks and what he *does not* speak in order to cooperate with the processes of signification. The expression of such processes - which can be both verbal (oral and written: intralinguistic processes) and alternative (gestures, body language, relational: intersemiotic processes) - go beyond difficulties and point to an individual's linguistic-cognitive efforts over their utterances.[11]

Finally, in episode 4, three years after the diagnosis, the progression of the disease showed an increase in the use of gestures for production of utterances. The use of repetition and specularity as a discursive mechanism is decreased, and gestures begins to serve as intersemiotic translation[11], either keeping up with speech (turn 16) or replacing speech (turns 12 and 14). In this context, the individual can express himself by constructing meaning from a verbal sign into a non-verbal sign, as occurs in turns 12 and 14, in which Heitor points to the window instead of referring to that he can see through it [11].

Through this semiotic inter-relationship, it can be seen that Heitor understands what his interlocutor says (turn 5 - "*Don´t you ever take the bus?"*), although his answer is gestural (turn 6 - ((reaches for his wallet and fetches his bus pass)) when he attempts to answer the question affirmatively. In other words, the adoption of alternative signification strategies, in the face of verbal difficulties, suggests that, at this stage of the disease, the individual's enunciative intention and comprehension are both relatively preserved. In this scenario, to effectively understand what Heitor means, the interlocutor has to make various interpretation efforts, which involve considering different semiotic channels, different routes for construction of sense and meaning, as well as isolated gestures, as if those enunciated by Heitor (verbal and non-verbal ones) were an "enigma" to be unveiled, a construction of meanings among the hypotheses of the interlocutor and the clues given by Heitor.

There are more frequent instances of pauses and hesitations (turns 4, 8, 10, 12, 18, 20), which clearly show the progression of linguistic difficulties already reported in previous episodes; however, they also signal moments when Hector is working on access and lexical selection and organizing his utterances[9,11,14]. In other words, the occurrences of dysfluencies, found (albeit at a lower frequency) also in normal speech, imply a stepback in verbal construction: one does not simply say just anything to fill in the gaps of silence; one hesitates, seeks, questions, feels puzzled, resorts to the interlocutor about what needs to be said next. At the moments of dysfluency, interlocutor and individual work collaboratively for the purpose of enunciation by negotiating meaning with the aim of ensuring the fluency of conversation and dialog. This concept of language as joint work to construct meaning and maintain fluency refers to the concept of *active-responsive act*, in which understanding is not merely a matter of decryption, but it is also an action by the one who listens[15], in this case, the interlocutor therapist. In this way, the interlocutor plays a central role in the dialog by managing, leading, collaborating and, finally, by promoting the implementation of the discursive intention of the individual [11.15].

FINAL REMARKS

This study showed that the analysis of language of an FTD patient, in the light of an enunciative-discursive approach from Neurolinguistics, ultimately highlights not only the deficits that occur in the disease - which have been previously described in the literature - but also, and above all, the patient's efforts to produce language, which are noticeable even when language is changed. Such efforts change as the disease progresses; they resort to verbal strategies and other semiotic channels (e.g. gestures). Moreover, they indicate that there is language in use, although under new rules. Therefore, one reaffirms the endeavor of overcoming the dichotomies that involve language and cognition when attempting to understand language pathologies. Language work is always shared, collaborative work between interlocutors involved in the dialog for construction of meaning. In this regard, interlocutors play a major role as they work increasingly towards developing active-responsive understanding. Thus, the enunciative-discursive analysis of dialog goes beyond the concern about how much an individual has or has not understood a question asked, or how much worse his language skills may become along the process of degeneration. Instead, therapy-oriented research should focus on the means that can promote the construction of the individual's intended utterances and help them maintain their role even when their speech is virtually absent. In other words, therapists, when taking their position in a privileged place of interlocution, should recognize both a language in operation and an individual who, in spite of his language-cognition deficits, can still manage to take his role as a "speaker".

In short, it is concluded that the analysis of dialog of language used by FTD patients allows us to understand the changes in language during the progression of the disease, as well as the mechanisms developed for the maintenance of his ability to express himself, thus providing evidence of a communicative intent of an FTD patient even at a more advanced stage of the disease. It can be concluded that the individual's expression of meaning is increasingly inter-related to the collaboration of the interlocutor in the construction of meaning, to active understanding of what an FTD patient enunciates, either through verbal signs or not. Thus, one can gain a wider perspective about the individual's relevant verbal production and deeper understanding of language production in the context of the conditions of verbal production, the interlocutor and the individual's life history - elements which compose the enunciative scene of such production.

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Author contributions

DMU participated in the design of the study, the review of the literature, data collection and analysis, and also wrote the manuscript; KPS participated in the review of the literature, discussion, revision and writing of the manuscript; APOS participated in the design of the study, organization of timetable, data collection and analysis, the review of the literature, discussion and final review of the manuscript.