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Bilingualism in a Case of the Non-fluent/agrammatic Variant of Primary Progressive Aphasia

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There is a growing body of research on language impairment in bilingual speakers with neurodegenerative diseases. Evidence as to which language is better preserved is rather inconclusive. Various factors seem to influence language performance, most notably age of acquisition, level of proficiency, immersion and degree of exposure to each language. The present study examined fluency, lexical, discourse and grammatical abilities of a Greek-French late bilingual man with the non-fluent/agrammatic variant of primary progressive aphasia (nfvPPA). Speech samples derived from three different narrative tasks in both languages were analyzed using quantitative production analysis (QPA) and fluency measures. The first aim of the study was to compare the participant's connected speech production to that of Greek-speaking normal controls. The second aim was to determine whether Greek (L1) and French (L2) were differentially impaired. To our knowledge, this is the first report of connected speech deficits in a Greek-speaking patient with PPA and the first study which uses QPA to compare L1 and L2 narratives in a bilingual speaker with PPA. Compared to neurologically healthy controls, our participant was impaired in lexical, discourse and grammatical productivity measures, but did not differ in measures of grammatical accuracy. The presence of dysfluencies, reduced speech rate and simplified syntax is consistent with the pattern of impairment reported for the nfvPPA. Results showed that narrative production measures did not differ significantly between languages. However, they suggest a slightly worse performance in his second, non-dominant, language despite a similar pattern of impairment in both languages. Lengthy exposure to L2 and regular activation of L2 through daily use may explain the preservation of discourse abilities in his non-dominant language. This study calls attention to factors such as language dominance, proficiency, patterns of use, and exposure to a language. These factors play a key role in assessing bilingual individuals with PPA and making clinical decisions.

Keywords: bilingualism, primary progressive aphasia, PPA, non-fluent, Greek, quantitative production analysis, connected speech, narrative

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INTRODUCTION

The notion of bilingualism refers to the use of two or more languages by an individual in daily life (Grosjean, 1994). First language (L1) and second language (L2) are typically the terms used to characterize languages in respect to their order of acquisition. The terms early and late bilingual classify a person according to the age at which the second language is acquired. Finally, the terms dominant and non-dominant language refer to differences in processing abilities between the two languages and/or in language use. Most researchers agree that both proficiency and use are key contributors to the bilingual experience (Treffers-Daller, 2015).

Bilingualism is a complex construct. Various factors seem to influence language performance in bilingual individuals. Factors related to L2, include age of acquisition, method of acquisition, level of proficiency in the second language and in different modalities (listening, speaking, reading, and writing), similarity to the first language and patterns of language use (e.g., Lorenzen and Murray, 2008; Goral and Conner, 2013; Kambanaros, 2016). In bilingual speakers with an acquired language disorder, language performance in L1 and L2 also depends on the underlying pathophysiology including traumatic brain injury, stroke and neurodegeneration.

Different hypotheses have been put forward to account for language representation in the brain. Evidence comes from electrophysiological investigations and neuroimaging studies of impaired and unimpaired bilingual persons, as well as clinical studies examining the effect of brain damage on language processing in bilingual speakers.

In terms of lexical processing, clinical studies support non-selective lexical access to a multilingual lexicon with shared lexical-semantic representations (e.g., Abutalebi, 2008; Kambanaros, 2016). Parallel lexical-semantic decline in cases of neurodegeneration (Hernández et al., 2008; Costa et al., 2012) or impairment in post-stroke aphasia (Kambanaros and van Steenbrugge, 2006; Kambanaros, 2009, 2010, 2016; Faroqi-Shah and Waked, 2010) are in favor of a common underlying neural network. Neuroimaging studies indicate both shared and separated brain regions for the two languages (Khachatryan et al., 2016).

As for grammar processing, researchers (Paradis, 1994, 2008; Ullman, 2001) have proposed that L1 and L2 are differentially processed as they rely on different cognitive mechanisms: L1 is acquired implicitly through immersion, whereas L2, when it is acquired later in life, explicitly through tuition. Syntactic processes are served by different brain areas, more left anterior (frontal) and subcortical (basal ganglia) regions for L1 and more posterior (temporo-parietal) cortical regions for L2. Others support shared L1 and L2 grammatical representations which are located in common regions (Hartsuiker et al., 2004; Weber and Indefrey, 2009). Evidence from functional neuroimaging studies suggest that L2 processing may become more automatic and converge to the same neural representations of L1 through long exposure to L2 (Abutalebi, 2008). However, differences between first and second language processing have been attributed to cognitive control mechanisms, as the functional demand placed on these regions is higher for speakers of multiple languages and influenced by factors such as age of acquisition, level of proficiency, and exposure to a language (Abutalebi and Green, 2007; Green and Abutalebi, 2013; Weber et al., 2016).

Evidence from brain imaging studies emphasize the role of L2 proficiency and age of acquisition in interpreting results. In studies where the level of proficiency has been controlled for, there is a higher degree of L1 and L2 overlapping activation for high-proficient than for low-proficient participants (Higby et al., 2013). The dorsolateral prefrontal cortex, anterior cingulate cortex, and right inferior frontal gyrus have been associated with L2 processing in lower proficient bilinguals in a meta-analysis by Sebastian et al. (2011). In another meta-analysis examining the role of age of acquisition in L1 and L2 processing, Liu and Cao (2016) concluded that language networks are more divergent for late bilinguals than for early bilinguals. Regions that were found to be more involved in L2 than in L1 processing were left insula and left middle frontal, inferior frontal and precentral gyri. The left superior frontal gyrus was more recruited by late bilinguals. This result suggests reliance on wider neural resources in the case of late bilinguals.

Primary progressive aphasia (PPA) is a neurodegenerative disease in which language is selectively impaired, at least in the initial stages, providing thus a unique opportunity to study bilingual aphasia and brain representations of language (Filley et al., 2006; Machado et al., 2010). The present study sought to investigate the connected speech deficits in a Greek-French late bilingual person with the non-fluent/agrammatic variant of PPA (nfvPPA). The nfvPPA is characterized by agrammatic production and/or apraxia of speech. Object knowledge and single-word comprehension are usually spared, whereas syntactic comprehension may be impaired. According to the 2011 consensus criteria (Gorno-Tempini et al., 2011), PPA also comprises the semantic (svPPA) and the logopenic (lvPPA) variant. Recently, primary progressive apraxia of speech (PPAOS) has been recognized as a distinct clinical entity (e.g., Duffy et al., 2014). Individuals with PPAOS present with apraxia of speech as their primary deficit and have little or no evidence of aphasia.

Single word production deficits have been extensively examined in PPA and studies of bilingualism. However, connected speech analysis has only recently begun to be systematically studied and has been used only in one study to compare performance in bilingual speakers with PPA (Zanini et al., 2011). The evaluation of connected speech enables a multi-level naturalistic assessment of language production (Marini et al., 2011). All linguistic levels, phonetics, phonology, morphology, syntax, semantics, pragmatics, and discourse can be evaluated when analyzing connected speech samples. Different tasks have been used to elicit speech samples and evidence suggests that they have different specificity for addressing different linguistic levels (Boschi et al., 2017). For example, a picture description task may be more useful in documenting lexico-semantic deficits, whereas story narration tasks favor the evaluation of discourse and syntactic abilities. Spontaneous speech production tasks are more sensitive to morphological, syntactic, and discourse level deficits, as in unconstrained tasks it is easier for speakers to compensate for their word-finding difficulties.

Deficits in the nfvPPA can arise at the phonetic-phonological level and manifest as a motor speech impairment and/or at the lexical-semantic, morphosyntactic, syntactic, or discourse level and present as agrammatism. Boschi et al. (2017) reviewed the evidence from studies focusing on connected speech deficits in neurodegenerative disorders. People with the non-fluent/agrammatic variant of PPA typically speak at a slower speech rate than healthy controls and make frequent speech sound errors (Ash et al., 2009; Wilson et al., 2010; Rogalski et al., 2011). At the lexical level, an increased number of errors in closed class words has been reported (Knibb et al., 2009; Meteyard and Patterson, 2009; Sajjadi et al., 2012). At the syntactic level, they make grammatical errors (Graham et al., 2004; Sajjadi et al., 2012) and produce simplified sentences with lower number of words per utterance, clauses, verb phrases, and coordinated sentences (Knibb et al., 2009; Wilson et al., 2010; Fraser et al., 2014). Concerning discourse abilities, individuals with the nfvPPA produce a reduced number of words, limited relevant information and they have difficulty maintaining the topic (Graham et al., 2004; Wilson et al., 2010; Sajjadi et al., 2012; Ash et al., 2013; Fraser et al., 2014).

Apart from allowing a multi-level evaluation of the speech and language deficits observed in PPA, connected speech measures enable comparison of patterns of impairment in different languages. For these reasons connected speech analysis has been deemed appropriate for the evaluation of narrative production in our bilingual subject with the nfvPPA. For the structural analysis of connected speech, we used the Quantitative Production Analysis (QPA) (Saffran et al., 1989). QPA was first used to describe agrammatic speech but has been found useful in identifying differences between fluent and non-fluent types of aphasia (e.g., Varkanitsa, 2012) and has been successfully applied in distinguishing normal from aphasic production and differentially diagnosing PPA variants (Wilson et al., 2010). An additional set of fluency measures, error analysis and macrolinguistic measures were also used to allow for a more thorough documentation of the deficits observed in nfvPPA.

A small number of case studies on bilingual speakers with PPA have been published in recent years (Filley et al., 2006; Hernández et al., 2008; Machado et al., 2010; Zanini et al., 2011; Larner, 2012; Druks and Weekes, 2013). Kambanaros and Grohmann (2012) published a case study of a multilingual man with fluent PPA, highly proficient in three languages, Greek, English, and Czech. He was more impaired in L3 than L2 and L1, and more impaired in L2 than in L1. In other words, the extent of impairment in each language was correlated with the order of acquisition. In a short report Machado et al. (2010) presented a Portuguese–French bilingual speaker with PPA. He was impaired in both languages. Performance was overwhelmingly better in his L1 which was also his dominant language. Larner (2012) in another short report, described a Welsh-English speaker who used her L1 in daily communication although L2 was her dominant language. In a more detailed study, Hernández et al. (2008) presented a Spanish-Catalan early bilingual individual with nfvPPA. They found a naming deficit which was more pronounced for L2 than for L1 at first assessment, but a parallel pattern of decline in both languages, even though L2 deteriorated more rapidly. A grammatical category-specific deficit was present in both languages with an advantage in noun naming over verb naming. A Hungarian-English late bilingual speaker with nfvPPA was reported by Druks and Weekes (2013). Their participant was more impaired in L2 which was his dominant language. A parallel deterioration was found for lexical and grammatical knowledge in L1 and L2. Zanini et al. (2011) described a case of an early Friulian-Italian bilingual woman with nfvPPA. They analyzed her spontaneous speech production and found more phonemic paraphasias, morphological and syntactic errors in L2 than in L1. They reported similar scores for number of dysfluencies, discourse productivity, grammatical productivity, and lexical selection measures (i.e., total words, utterances, subordinate clauses and open-class words) in both languages. Only Filley et al. (2006), who presented a Chinese–English-speaking woman with the logopenic variant of PPA, have reported a non-significant better performance for repetition, naming and conversation tasks, but more phonemic paraphasias, in L2 which was her dominant premorbid language. A parallel pattern of deterioration was observed in both languages. To conclude, most of these studies have found evidence of greater impairment in L2, irrespectively of language dominance and age of acquisition, indicating that L2 may be more vulnerable to degeneration than L1.

In the context of neurodegenerative diseases, there is also a growing body of group studies on language impairment in bilingual speakers with Alzheimer's Disease (AD). The available evidence is mixed. Some studies report parallel deterioration (Salvatierra et al., 2007; Costa et al., 2012; Manchon et al., 2015; Nanchen et al., 2017), while others report differential deterioration of the two languages (Mendez et al., 1999; Gollan et al., 2010). In the study by Gollan et al. (2010), bilingual persons with AD exhibited greater decline in the dominant than the non-dominant language. An opposite pattern was found by Mendez et al. (1999). Based on caregivers' reports, they concluded that the non-dominant language was more affected than the dominant language. Ivanova et al. (2014) found different longitudinal and cross-sectional patterns of decline. The non-dominant language declined more than the dominant language, but differences between patients and controls were greater for the dominant than for the non-dominant language. The authors concluded that both languages are affected by AD with different trajectories of decline over time.

The aim of the present study was 2-fold. First, to provide an account of connected speech deficits in the non-fluent variant of PPA in Greek. The participant's speech and language deficits in his native language were examined by comparing performance on connected speech elicited from a picture description task with speech samples obtained from a healthy control group on the same task. Second, to compare performance in Greek and French and evaluate impairment patterns in both languages connected speech samples from three different narrative tasks in each language were elicited. To our knowledge, this is the first report of connected speech deficits in a Greek-speaking patient with PPA and the first study which uses QPA to compare L1 and L2 narratives in a bilingual speaker with PPA.

The two languages differ in several respects. Greek is classified as an independent branch within the family of Indo-European languages, whereas French belongs to the Romance branch of the Indo-European family. The components of morphology and syntax are especially relevant to our study. Subject-verb-object (SVO) order is the basic word order in both languages. Word order is flexible in Greek, whereas French has a relatively strict word order. Moreover, Greek is a null subject language, i.e., subjects are not typically expressed when they can be inferred from the context (Roberts and Holmberg, 2010). On the other hand, French is a non-null subject language which requires an explicit subject in a sentence. Regarding morphology, Greek is a highly inflected language, whereas French is considered to be a moderately inflected language. The main difference between the two languages is that in Greek nouns, pronouns, and adjectives are inflected not only for number and gender but also for case. Case in French is expressed using mainly word order and prepositions (Prévost, 2009), although there is a morphological case marking system for weak object pronouns (clitics).

Despite the different linguistic properties of Greek and French, which may result in differences in the narrative measures (e.g., higher proportion of pronouns in French than in Greek because of the mandatory inclusion of subjects in sentences), we predict a similar pattern of impairment in both languages. We also predict that L2, the participant's non-dominant and less proficient language, will be affected to a greater degree compared to L1.

MATERIALS AND METHODS

Participant

Participant LJ is a chef in his early sixties, with 6 years of formal education. He is a right-handed late bilingual whose native language (L1) is Greek. At the age of 25, he moved to a French-speaking country and worked as a cook in a French-speaking environment for 7 years. On his return to Greece, he continued to use French (L2) both at work and at home with his wife who is a French native speaker. Details about his language history and proficiency were collected from his wife upon completion of the French version of the Language Experience and Proficiency Questionnaire (Marian et al., 2007) (**Table 1**). Language dominance was determined based on the reported proficiency and extent of language exposure. Task specific measures of proficiency (for understanding, speaking and reading), across settings measures of language exposure (to family, friend, reading and television) and global measures of these two dimensions were all taken into account in order to ascertain language dominance.

LJ reported a progressive deterioration of speech and language functions. Language impairment was the primary impairment for at least the first two years. LJ was initially assessed 5 years after symptom onset. He received a comprehensive evaluation including case history, neurological examination, and neuropsychological testing coordinated by the second author who is a psychiatrist specialized in memory disorders with extensive experience working with patients with degenerative diseases. He was referred for speech and language evaluation and completed an initial language assessment performed by the first author in Greek. He was diagnosed with PPA, as neuroimaging results ruled out other causes of focal brain damage and extensive white matter disease (see **Figure 1**) and was given a clinical diagnosis of non-fluent/agrammatic PPA according to current criteria (Gorno-Tempini et al., 2011). There

TABLE 1 | Reported language history and proficiency for participant LJ based on the Language Experience and Proficiency Questionnaire (LEAR-Q, Marian et al., 2007).

Language history measures Languages	L1 history Greek	L2 history French	L3 history English	Range
Order of proficiency	1	2	3	
Order of acquisition	1	3	2	
Identification with culture[a]	10	6	1	0–10
Current exposure	46%	46%	8%	
Preference for reading	80%	20%		
Preference for conversing	40%	40%	20%	
REPORTED PROFICIENCY[b]				
Understanding	4	5		0–10
Speaking	5	5		0–10
Reading	2	0		0–10
AGE MILESTONES (YEARS)				
Started learning		25		
Attained fluency		29		
Started reading	6	25		
Became fluent reading		n/a		
IMMERSION DURATION (YEARS)				
Country	53	7		
Family	53	36		
School/Job	53	36		
CONTRIBUTION TO LANGUAGE LEARNING[c]				
From family	10	10		0–10
From friends	0	8		0–10
From reading	0	0		0–10
From TV	2	5		0–10
From radio	0	0		0–10
From self -instruction	0	1		0–10
EXTENT OF LANGUAGE EXPOSURE[d]				
To family	10	10		0–10
To friends	10	7		0–10
To reading	1	1		0–10
To TV	7	3		0–10
To radio	0	0		0–10
Self -instruction	0	0		0–10
SELF -REPORTED FOREIGN ACCENT[e]				
Perceived by informant	2	5		0–10
Identified by others	5	5		0–10

[a]Range, 0 (none) to 10 (complete); [b]Range, 0 (none) to 10 (perfect); [c]Range, 0 (not a contributor) to 10 (most important contributor); [d]Range, 1 (never) to 10 (always); [e]Range, 0 (none) to 10 (pervasive).

were no signs of limb apraxia, tremor, dystonia and myoclonus. There was a very mild hypertonicity on the right side, as well as reports of becoming more suspicious of others. His speech was slow with word finding problems, hesitations, pauses, and sound errors. Motor speech evaluation determined the presence of apraxia of speech with slow overall rate, deliberate, slowly sequenced speech sequential motion rates in comparison to speech alternate motion rates, imprecise articulation with sound distortions, a tendency to equalize stress across syllables, false starts and restarts and sound and syllable repetitions. Dysarthria, most probably spastic, was present, but less severe than apraxia of speech. LJ had spared knowledge of objects and word recognition. A mild difficulty comprehending syntactically complex sentences was revealed in formal testing. His consensus score on the Progressive Aphasia Severity Scale (PASS) (Sapolsky et al., 2010) was 7 (see **Table 2**). Background linguistic and neuropsychological evaluation results are presented in **Table 3**.

Prior to testing for the present study, LJ had received speech and language therapy for approximately 4 months. Intervention included partner education, script training (Youmans et al., 2005) of telephone conversations with clients and techniques based on the "Oral Reading for Language in Aphasia" treatment program (Cherney, 2010) that addressed production of multisyllabic words, as well as reading and auditory comprehension. Treatment was delivered in Greek.

The present study was conducted 9 months after the initial evaluation (5 years and 9 months after the reported onset of the disease) and 3 months after the last therapy session. At the time of the study, LJ had a FTLD-modified CDR sum of boxes score of 9 (MMSE = 17/30). The Montreal Cognitive Assessment (MOCA) was administered both in Greek and French. He received a score of 18/30 in Greek and 20/30 in French (one additional point in visuospatial/executive function and one in memory). He generated 2 words in the phonemic verbal fluency task and 5 words in the semantic task (animals) and obtained a score of 3 on the forward digit span and 0 on the backward digit span. There was also a parallel deterioration of motor skills. These results suggest a deterioration in cognitive function, especially in the domain of executive function and progression of the nfvPPA to a corticobasal syndrome. Corticobasal syndrome can overlap clinically and pathologically with PPA and many cases initially classified as nfvPPA, meet the criteria for corticobasal syndrome at a later

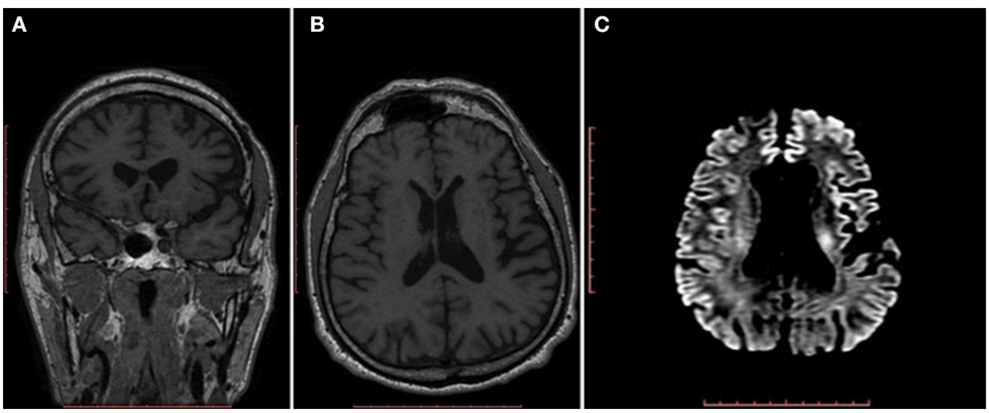

FIGURE 1 | Coronal T1-weighted **(A)**, axial T1-weighted **(B)**, and axial diffusion-weighted **(C)** brain imaging at initial assessment showing left perisylvian atrophy.

TABLE 2 | Consensus score on the Progressive Aphasia Severity Scale (PASS) at initial evaluation.

PASS Domains	Normal	Quest/ble very mild	Mild	Moderate	Severe
	0	0.5	1	2	3
Articulation			1		
Fluency			1		
Syntax and grammar			1		
Word retrieval - expression			1		
Repetition	0				
Auditory comprehension		0.5			
Single word comprehension	0				
Reading		0.5			
Writing			1		
Functional communication			1		

Severity (Sum of boxes), 7.

TABLE 3 | Background neuropsychological assessment results.

Area of testing and tests	Score (correct)
GENERAL COGNITIVE MEASURES	
MMSE	28/30
ACE-R	86/100
Attention	18/18
Memory	26/26
Fluency	5/14 *
Language	25/26
Visuospatial abilities	12/16 *
EXECUTIVE FUNCTIONING	
Frontal Assessment Battery (FAB)	12/18
VISUOSPATIAL PERCEPTION	
Benson figure test—Copy condition	15/17
VISUAL MEMORY	
Benson figure test—Delayed recall condition	17/17
MOOD	
GDS-SF	3/15
IDEOMOTOR APRAXIA	
WAB	58/60
REPETITION	
Informal (based on WAB)	95/100
NAMING	
Boston Naming Test, BNT-SF	11/15 *
LANGUAGE COMPREHENSION	
Vocabulary (PPVT-32)	19/32
Auditory comprehension-words (BDAE-SF)	16/16
Sequential commands (BDAE-SF)	10/10
Written sentences/passages (BDAE-SF)	4/4
Written story (BDAE-SF)	3/3
Grammaticality judgment—morphology (Fyndanis et al., 2013)	77/80
Syntactic comprehension (BDAE-3)	8/10
OBJECT SEMANTICS	
Pictures (PPTT-SF)	14/14
READING EFFICIENCY (Simos et al., 2013)	
Real words	16 in 45s *
Pseudowords	13 in 45s
WRITING	
Words (Informal)	7/20
Non-words (Informal)	14/14
Words (BDAE-SF)	8/9
Written picture description (BDAE-SF)	4/11 *
MOTOR SPEECH EVALUATION (Wertz et al., 1984)	
Apraxia of speech rating	3/7
Dysarthria rating	1/7

**Significant impairment (>2 standard deviations below the normative mean); MMSE, Mini Mental State Examination (Fountoulakis et al., 2000); ACE-R, Addenbrooke's Cognitive Examination-Revised (Konstantinopoulou et al., 2011); GDS-SF, Geriatric Depression Scale-Short Form (Fountoulakis et al., 1999); WAB, Western Aphasia Battery; BDAE-SF, Boston Diagnostic Aphasia Examination Short form (Goodglass et al., 2013); PPVT, Peabody Picture Vocabulary Test (Simos et al., 2011); PPTT-SF, Pyramid and Palm Trees Test-Short Form (Breining et al., 2015).*

stage (Grossman, 2010; Duffy et al., 2014; Leyton and Ballard, 2016; Santos-Santos et al., 2016).

The study was approved by the ethics committee of the Athens Alzheimer's Association. The research was conducted in accordance with the latest version of the Declaration of Helsinki. LJ was informed about the purpose and procedures of the study and gave written consent for participating in the study, as well as for the recording and publication of his clinical data. Both LJ and his wife gave written informed consent for the publication of this manuscript. The initials LJ are fictional.

Elicitation and Transcription of Speech Samples in L1 (Greek) and L2 (French)

Three different speech samples were collected in both Greek and French, under 3 conditions: a picture description task ("cookie theft," from Boston Diagnostic Aphasia Examination, BDAE), a story retell task (the dog story protocol from the Multilingual Assessment Instrument for Narratives, MAIN, Gagarina et al., 2012, 2015) and a semi-spontaneous speech task where LJ was asked to talk about his job. Interruptions and questions by the examiner (first author) were kept to a minimum. The examiner is a monolingual Greek-speaking clinician who is also a proficient speaker of French. Samples were collected in 4 sessions, first for the Greek language and 2 weeks later for French. All samples were audio-recorded.

Speech samples were transcribed orthographically using ELAN (Sloetjes and Wittenburg, 2008). Phonological paraphasias unintelligible or incomprehensible words were transcribed phonetically using the International Phonetic Alphabet. Dysfluent variables, such as silent and filled pauses, sound errors, repetitions, and false starts were also coded.

Quantitative Analysis of Speech Samples

Speech samples were analyzed following the procedures described by Saffran et al. (1989) for quantitative production analysis (QPA) (Saffran et al., 1989; Berndt et al., 2000; Rochon et al., 2000). The QPA procedures were followed for all samples, with the exception of the direct discourse utterances produced in the story retell task, which contrary to the QPA instructions were not excluded, as these structures were modeled in story-telling. Narrative samples were formed by extracting comments on the narrative, direct responses to the examiner, repetitions of the examiner's utterances, stylistic and dysfluent repetitions, subsequently repaired utterances and discourse markers. The narrative samples were then segmented into utterances based on semantic, syntactic, and prosodic information. Utterances and narrative words were used in subsequent analysis.

The QPA summary measures were classified into four categories: discourse productivity, sentence productivity, grammatical accuracy, and lexical selection (Gordon, 2006). A set of additional measures were used to quantify dysfluent speech and narrative variables.

Speech Rate and Other Fluency Variables

Speech rate for each sample was calculated by dividing total completed words by sample duration in minutes. Samples were timed, and total time duration was computed by subtracting the examiner's interjections.

Pauses longer than 1 s were coded according to QPA instructions and counted for the calculation of the pause frequency measure. However, a threshold of 0.250 ms was used in the calculation of pause duration (De Jong and Bosker, 2013) and speaking time was calculated by subtracting silent pausing time from total time in order to control for the effect of pauses. Articulation rate was computed by dividing total completed words by speaking time.

Speech sound errors included distortions, which were defined as phonetic errors resulting in distorted phonemes, and phonological paraphasias defined as words with non-distorted phonemic insertions, deletions, or substitutions. Whole-word immediate repetitions were counted as dysfluent repetitions. Words or phrases repeated later in the narratives were counted as speech repairs. Partially produced words were coded as false stars and small words, such as "eh," as filled pauses.

Speech samples were of different duration and direct comparison of the aforementioned frequency measures was not possible. Thus, these measures were calculated as proportions of total words produced. They were also corrected for speaking length by dividing dysfluency counts by speaking time (De Jong, 2016).

Discourse Measures

QPA discourse productivity measures included speech rate, number of narrative words, and proportion of narrative to total words produced, as a measure of discourse efficiency.

An additional discourse variable, Guiraud's index (the square root variant of Type-Token Ratio, TTR) was also measured. Guiraud's index is a measure of lexical richness that is less affected by sample size/length in comparison to TTR (Van Hout and Vermeer, 2007). This was derived by dividing the number of unique words (types) by the square root of narrative words (tokens). Number of unique words (types), lemmas, and utterances are also reported.

Lexical Measures

Grammatical category class (closed/open class, nouns, verbs, adjectives, adverbs, pronouns, prepositions, conjunctions) was coded for each narrative word. Their proportion was calculated by dividing the number of words in each category by the number of narrative words. Nouns, verbs and adjectives were considered as open class. All other words were counted as closed class. Proportion of verbs to nouns and verbs was also computed. Proportion of pronouns was derived by dividing the number of pronouns by the total number of nouns and pronouns.

Finally, mean log word frequency of open class words was calculated for each narrative sample. Calculations were based on data about word frequencies per million taken from the "ILSP PsychoLinguistic Resource" for the Greek language (Protopapas et al., 2012) and "Lexique" for the French language (New et al., 2001).

Grammatical Measures

QPA sentence productivity measures encompass proportion of words in sentences, mean utterance length (in words), median utterance length (in words), sentence elaboration index (number of open class words per phrase for noun and verb phrases), and an embedding index (proportion of embeddings to sentences).

QPA grammatical accuracy measures consist of proportion of well-formed sentences, verb inflection index (proportion of inflectable verbs inflected) and determiner index (proportion of determiners produced in obligatory contexts). The auxiliary complexity index, a measure of morphological complexity of the main verb indicating change from its base form, was also calculated.

Macrolinguistic Analysis (MAIN)

Narrative assessment focused on the analysis of microlinguistic aspects of language production. Macrolinguistic aspects were addressed for the "Dog story" retell task with the story structure score and the structural complexity measures proposed by MAIN (Gagarina et al., 2012, 2015). Although the MAIN was originally designed to assess narrative skills of bilingual children, it is controlled for macro-and microlinguistic features across Greek and French. As there is no other standardized procedure for adults, it was deemed appropriate for comparing story retell abilities in both languages.

The "Dog story" starts with a setting statement and consists of three short episodes. Each episode consists of an initiation, a goal, an attempt, an outcome and a reaction statement. Credit is given for the production of each initiation, goal, outcome, reaction when computing the story structure score.

Five measures of structural complexity, included in the MAIN, were calculated: number of sequences where an attempt and outcome statement has been generated (but no goal), number of single goal statements, number of incomplete episodes which they include a goal and an attempt statement sequences, number of incomplete episodes which they include a goal and an outcome statement, and number of complete episodes which include all three goal-attempt-outcome components. Comprehension of the story structure was also assessed by means of questions targeting the main macrostructure components.

Error Analysis

The following type of errors were also identified and measured as a proportion of narrative words.

- Syntactic errors were recorded when LJ produced ungrammatical sentences.
- Morphological errors, affecting articles, nouns, adjectives, and verbs, were counted separately.
- Semantic errors included selections that were semantically inappropriate for the context.
- Code switching errors were defined as words produced in languages other than the target language (number of tokens not in the target language).

Some morphological errors in L2 (article-noun gender agreement) occurred with the same nouns. These persistent errors were not included in individual error counts but contributed to the calculation of the total number of errors.

Inter-rater Reliability

Analysis of 30% of the Greek speech samples was completed by 2 additional raters both native speakers of Greek with some linguistic training. Spoken word interrater reliability ranged from 90 to 95%. A consensus for each point of disagreement was reached through a discussion between the raters.

Control Group for QPA

QPA measures for the picture description task in Greek were compared to the measures of a control group included in a previous study by Varkanitsa (2012). Varkanitsa used the QPA protocol in order to compare the connected speech of Greek-speaking persons with aphasia following stroke to that of neurologically healthy adults. The same picture description task was used in the present study to elicit speech samples. Taking into account the fact that in Greek isolated verbs may constitute grammatical utterances, Varkanitsa categorized utterances as "utterances with verb," "utterances without verb" and "single-word utterances." The QPA protocol was applied without other modifications. The control group consisted of six normal native Greek speakers (3 males and 3 females) with a mean age of 61.17 (SD = 5) years and a mean of 9 (SD = 4.15) years of education.

There was no control group for QPA measures in French, as we did not have access to a French-speaking population and published studies, which have applied QPA in French-speaking individuals, have not used the same methodology. For this reason, our analysis focused on the pattern of deficits observed in the two languages. Moreover, careful consideration was given to cross-linguistic differences.

Statistical Analysis

LJ's narrative scores for the picture description task in Greek were compared to the scores of a neurologically healthy control group (Varkanitsa, 2012). T-values were calculated using Crawford and Howell's method which enables the comparison of performance of a single subject with that of a small control sample (Crawford and Garthwaite, 2012). Differences between LJ's performance in Greek (L1) and French (L2) were calculated using the Wilcoxon signed-rank non-parametric test for related samples because of the small sample size. Finally, scores from both languages were collapsed and correlations between errors and fluency, lexical productivity, grammatical accuracy, and productivity measures were calculated using the non-parametric Kendall's tau-b correlation coefficient due to the limited number of samples used in the analysis.

RESULTS

QPA Measures for the Picture Description Task in Greek—Comparison to Healthy Subjects

LJ's scores for the picture description narrative in Greek are presented in **Table 4**. His speech rate was slow, 40.37 words per minute. In the picture description task, he made two syntactic errors. Both errors involved the omission of obligatory post-verbal arguments. He also made speech errors. Dysfluencies included silent pauses, filled pauses, false starts, sound distortions, and repetitions (23, 20, 3, 2, and 1%, respectively, of total words produced). Compared to the control group, LJ used less narrative words [$t_{(5)} = -2.089$, $p < 0.05$] and more single word utterances [$t_{(5)} = 7.869$, $p < 0.0005$] to describe the picture. Sentence productivity measures (mean length of utterance, elaboration index and embedding index) did not differ from controls. LJ produced less nouns [$t_{(5)} = -2.468$, $p < 0.05$] and adverbs [$t_{(5)} = -3.240$, $p < 0.025$]. On the other hand, he produced more pronouns [$t_{(5)} = 7.406$, $p < 0.0005$] and verbs [$t_{(5)} = 2.546$, $p < 0.05$] than the control speakers.

TABLE 4 | LJ's scores, control group median and standard deviation values and Crawford-t values.

Spoken language measures	LJ	Controls[a] ($n = 6$) Median (SD)	t-values[b]
Proportion of closed class words	0.52	0.53 (0.04)	−0.23
Proportion of nouns	0.17*	0.25 (0.03)	−2.47
Proportion of adjectives	0.04	0.02 (0.01)	1.85
Proportion of prepositions	0.02	0.06 (0.02)	−1.85
Proportion of adverbs	0*	0.07 (0.02)	−3.24
Proportion of pronouns	0.14**	0.06 (0.01)	7.41
Proportion of verbs	0.31*	0.20 (0.04)	2.55
MLU	4	5.41 (1.08)	−1.21
Elaboration index	1	2.43 (1.43)	−0.93
Embedding index	0.36	0.36 (0.17)	0.00
Number of narrative words	48*	127 (35)	−2.09
Proportion of sentences	0.92	0.79 (0.11)	1.09
Proportion of utterances without verbs	0.08	0.19 (0.11)	−0.93
Proportion of single-word utterances	0.17**	0.00 (0.02)	7.87
Proportion of well-formed utterances	0.75	0.96 (0.65)	−0.30
Auxiliary complexity index	0.64	0.30 (0.27)	1.17

[a]*Control group values are taken from Varkanitsa (2012);* [b]*One-tailed (*$p < 0.05$; **$p < 0.01$).*

Comparison of L1 and L2

Statistical analysis using the Wilcoxon signed-rank test revealed that the connected speech measures used to quantify speech production in L1 and L2 did not differ significantly across languages.

Fluency Measures

The mean duration of narratives was 2.24 (SD = 0.09) minutes for L1 and 3.76 (SD = 1.86) for L2. Pause duration, for pauses >0.250 ms, was 0.74 (SD = 0.11) minutes for L1 and 1.27 (SD = 0.36) for L2. Speaking time was 1.5 (SD = 0.08) minutes for L1 and 2.49 (SD = 1.58) for L2. Speech rate was faster for L2 than for L1, 44.10 (SD = 5.96) and 38.24 (SD = 2.52) words per minute (wpm), respectively. Similar results were noted for articulation rate: 73.00 (SD = 19.09) wpm for L2 and 57.43 (SD = 6.93) wpm for L1. However, these differences were not statistically significant.

Dysfluencies included silent pauses, fillers, false starts, distortions and immediate repetitions of whole words and in particular closed class words. The different types of dysfluencies are presented in **Figure 2**.

Although differences between languages did not reach statistical significance, there is a trend toward making more repetitions in L2, 0.040 (SD = 0.012) than in L1, 0.004 (SD = 0.007).

Discourse Measures

LJ produced longer narratives in L2 than in L1, 94.67 (SD = 68.06) words and 16.33 (SD = 10.12) utterances vs. 53.00 (SD = 8.66) words and 11.00 (SD = 3.61) utterances, respectively. Differences were not significant. From the narrative words, 47.00 (SD = 11.36) words in French and 34.33 (SD = 3.22) words in Greek were unique. Proportion of narrative to total words produced was 0.61 (SD = 0.07) in L1 and 0.55 (SD = 0.21) in L2.

Lexical Measures

Regarding word class production, significant differences between L1 and L2 were not found. However, LJ produced more closed class words and pronouns in L2 compared to L1 narratives. In French, the proportion of closed class words was 0.56 (SD = 0.01), while in Greek, it was 0.49 (SD = 0.03). The proportion of pronouns was 0.22 (SD = 0.04) in L2, as opposed to 0.12 (SD = 0.04) in L1. LJ produced personal, demonstrative, indefinite and interrogative pronouns. In L1, all demonstrative pronouns (37.5%) were used as subjects, whereas all the rest, including personal pronouns (50%) in their weak form, were produced as object pronouns (62.5%). Of all the pronouns produced in L2, 87.7% were personal pronouns and 8.78% demonstrative. Ninety-Four percent of the personal pronouns were used in their strong form and the remaining 6% in their weak form. In L2, 87.7% of the pronouns produced were subject pronouns and 12.3% object pronouns.

LJ used more nouns per narrative words in Greek, ranging from 0.17 to 0.31 with a mean of 0.26 (SD = 0.08), in comparison to his L2 in which the proportion of nouns was 0.17 (SD = 0.04), ranging from 0.12 to 0.20. The proportion of verbs produced did not differ across languages (see **Table 5**).

LJ used more high frequency words in French than in Greek narratives. The mean logarithmic frequency of French open class words was 1.71 (SD = 0.17), as opposed to 1.392 (SD = 0.15) for Greek words. This difference was not statistically significant.

Grammatical Productivity and Accuracy Measures

With regard to measures associated with grammatical production, no statistically significant differences were found between L1 and L2. Mean and median length of utterance in words was 5.12 (SD = 1.53) and 4.17 (SD = 1.04) for Greek and 5.58 (SD = 0.59) and 5.00 (SD = 1.00) for French respectively. LJ performed more poorly in L2 than in L1 as far as the proportion of embedded clauses is concerned (0.19 (SD = 0.08) for L2 and 0.34 (SD = 0.17) for L1).

Macrolinguistic Measures for MAIN

The MAIN story structure and comprehension scores were 7/17 and 10/10 in L1 and 9/17 and 7/10 in L2, respectively. LJ produced one single goal statement in both languages. In French, he also used a sequence with an attempt and outcome statement. Neither incomplete episodes with a goal and an attempt/outcome statement nor complete episodes (with all three components) were present in his narratives.

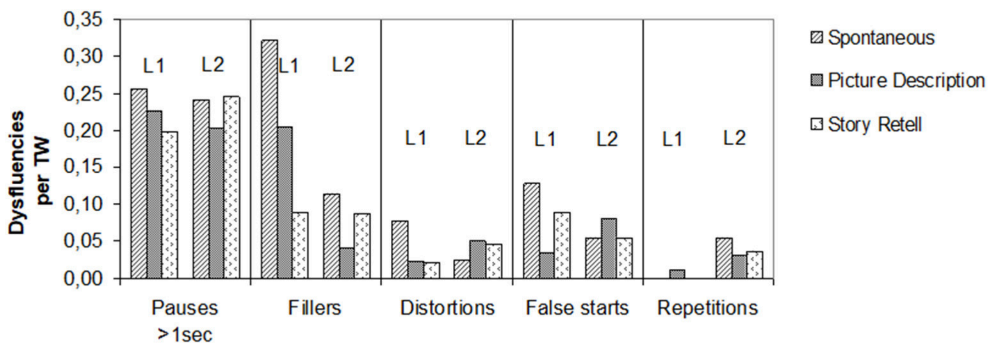

FIGURE 2 | Dysfluencies per total words (TW) in L1 and L2.

TABLE 5 | Proportion of nouns, verbs and pronouns per narrative words (NW) produced in personal narrative (task1), picture description (task 2) and story retell (task 3) in L1 and L2.

	L1				L2			
Proportion per NW	**Task 1**	**Task2**	**Task 3**	**Total Mean (SD)**	**Task1**	**Task 2**	**Task 3**	**Total Mean (SD)**
Nouns	0.31	0.17	0.29	0.26 (0.08)	0.18	0.12	0.20	0.17 (0.04)
Verbs	0.21	0.31	0.24	0.25 (0.05)	0.20	0.30	0.18	0.23 (0.07)
Pronouns	0.08	0.15	0.14	0.12 (0.04)	0.20	0.26	0.19	0.22 (0.04)

Error Analysis

Systematic errors involving article gender agreement in L2 were excluded from analysis.

LJ made more morphological and semantic errors per narrative words in L2, 0.031 (SD = 0.025) and 0.022 (SD = 0.006), respectively, than in L1, 0.014 (SD = 0.024) and 0.005 (SD = 0.009), respectively. These differences were not statistically significant. Syntactic errors were stable across languages, 0.026 (SD = 0.016) for L1 and 0.023 (SD = 0.008) for L2.

Code switching was evident in one speech sample (spontaneous narrative) in French. LJ produced 11 out of the 166 complete words in Greek.

Correlational Analysis

We undertook correlational analyses between errors and connected speech measures. Syntactic errors were significantly correlated with the total number of dysfluencies per total words ($\tau b = 0.733$, $p = 0.039$), whereas morphological errors with the distortions produced per articulation minute ($\tau b = 0.966$, $p = 0.007$). Finally, there was a positive correlation between semantic errors and number of complete words ($\tau b = 0.867$, $p = 0.015$).

DISCUSSION

The present study examined fluency, lexical content, discourse and the grammatical abilities of a Greek-French late bilingual man with non-fluent/agrammatic PPA by analyzing speech samples derived from three different discourse tasks in both languages.

The first aim of the study was to compare the participant's performance to normal controls in L1. Compared to Greek-speaking neurologically healthy individuals, LJ was impaired in discourse and grammatical productivity measures, but did not differ in measures of grammatical accuracy. At the lexical level, there were some significant differences in the proportion of grammatical class words produced. In particular, LJ produced more verbs and pronouns, but less nouns and adverbs. However, proportion of closed class words was normal.

The second aim of the study was to determine whether or not L1 and L2 were differentially impaired. Results showed that discourse production measures did not differ significantly between languages. These findings indicate that both languages were similarly affected.

Comparison With Healthy Controls in L1 (Greek)

LJ produced a smaller number of narrative words, shorter utterances and simplified sentences compared to controls, as indicated by the MLU, proportion of single-word utterances and elaboration index measures. Production of embedded clauses was at the same level with the control group. The auxiliary complexity index, a measure of verb morphological complexity, was slightly higher for LJ than controls. However, the proportion of single-word utterances is the only grammatical productivity measure that reached statistical significance. Grammatical accuracy did not differ between LJ and neurologically healthy individuals, even though he produced a lower proportion of well-formed utterances. In the picture description task in Greek, LJ made two errors. Both errors were syntactic in nature and involved the omission of obligatory post-verbal arguments. Taken together, these results indicate an impairment at the discourse and grammatical productivity levels.

Fluency, as measured by speech rate and frequency of dysfluent errors, is another area that was affected. Although we had no control data for the fluency variables, slow speech rate and high proportion of pauses and fillers corroborate reduced fluency. Indicatively, a normal speech rate of 143.70 (SD = 23.40) wpm has been reported for the "cookie theft" description task in a study by Fyndanis et al. (2013). The measure was based on three neurotypical Greek-speakers with a mean age of 58 (SD = 9.64) years. The presence of distortions and false starts indicate an underlying motor speech problem, apraxia of speech in particular (Ogar et al., 2007; Wilson et al., 2010).

Differential impairment of nouns and verbs has been reported in aphasia resulting from stroke and PPA. In particular, disproportionate impairment of naming actions is commonly associated with non-fluent types of aphasia (Kambanaros, 2010) and greater verb naming impairment has been found in nfvPPA (Hillis et al., 2006; Ash et al., 2009; Thompson et al., 2012). Even though LJ used more verbs than nouns during the picture description task in Greek, indicating an opposite pattern of noun-verb dissociation, mean noun-verb ratio from all three Greek speech samples was within normal limits. In fact, higher proportion of verbs seems to be task-related, as disproportionate production of verbs was evident in both languages for the picture description task only. Normal ratios of nouns to verbs in connected speech of individuals with the nfvPPA have been reported in several studies (Graham et al., 2004; Knibb et al., 2009; Meteyard and Patterson, 2009; Fraser et al., 2013; Marcotte et al., 2017).

LJ also used more pronouns in Greek (0.14 per narrative words, 80% demonstrative, 20% personal) than the control group in the picture description task. Increased proportion of pronouns has been found in svPPA and it has been suggested that it may indicate lexical retrieval deficits, vague, or non-specific speech (Kavé et al., 2007; Meteyard and Patterson, 2009; Wilson et al., 2010; Fraser et al., 2014). Nevertheless, all the pronouns used by LJ had clear referents. Furthermore, all the demonstrative pronouns were used in the subject position of sentences. In a null subject language like Greek, demonstrative pronouns may be used as subjects to place additional emphasis on the referent. The production of overt subjects in Greek could reflect the influence of the syntactic properties of the participant's L2 on his L1. Syntactic attrition effects have been reported in the production of preverbal subjects in a group of Greek (L1) speakers, highly proficient in English (L2) (Tsimpli et al., 2004). However, in the personal monolog LJ produced a substantially lower proportion of pronouns (0.08 per narrative words) than in the two picture-based tasks. This most probably suggests that LJ was using demonstrative pronouns to direct the attentional focus to the referent in the depicted scenes. It must be noted that, although the examiner's instruction for the picture description task was "tell me everything you see going on in this picture," for the story retell task, the instructions focused on the story itself, not the pictures ("Can you tell me the story?" "Tell me more."). Picture-based tasks have been reported to result in the production of descriptions of the depicted items, rather than narrative samples (Bryant et al., 2016).

Wilson et al. (2010) used a similar methodology to ours by combining QPA and fluency measures to analyze narrative production of 50 English-speaking individuals with PPA. Speech samples were elicited through a picture description task. They found that their nfvPPA group compared to normal controls spoke slower, produced less words and their samples were of longer duration. All nfvPPA participants made distortions and more filled pauses than controls. Their mean length of utterances and number of embeddings were significantly reduced. In respect to the other variants of PPA, the authors concluded that the presence of distortions was the most informative measure for distinguishing between the nfvPPA and lvPPA. Additional measures that may assist in differentially diagnosing these subtypes are proportion of verbs and number of embeddings, which are higher in the lvPPA. Faster speech rate, less distortions, higher proportion of pronouns and verbs and nouns of higher frequency were found in the svPPA compared to the nfvPPA.

LJ's scores support the pattern of impairment reported for the nfvPPA variety. In comparison to neurotypical controls, he made distortions, spoke slower, produced less words and more single word utterances. Although agrammatism has been described as a core characteristic of this variant (Ash et al., 2009; Thompson et al., 2012), grammatical deficits may not be the primary feature of nfvPPA (Graham et al., 2004; Patterson et al., 2006; Wilson et al., 2010). In a recent study, Graham et al. (2016) evaluated fluency and grammatical production in nine individuals with nfvPPA. They reported that frank agrammatism was not always present and reviewing the literature they pointed out that grammatical abilities in persons with the nfvPPA show a high degree of variability. Nevertheless, researchers have consistently reported reduced speech rate, as well as simplified syntax and shorter utterances in connected speech in comparison to healthy controls (Ash et al., 2009, 2010, 2013; Knibb et al., 2009; Wilson et al., 2010; Marcotte et al., 2017).

Comparison of L1 and L2

The observed differences between L1 and L2 did not reach statistical significance, contrary to our hypothesis. This may be due to the small sample size of linguistic data or the between-task variability. Alternatively, findings may be interpreted as indicating a similar degree of impairment in both languages. Before commenting on this finding, there are some trends in the results that are worth mentioning.

The total number of dysfluencies was similar across languages. However, LJ produced more immediate repetitions in L2 than in L1. He repeated mostly personal pronouns at the beginning of utterances, or after silent pauses. In French, personal pronouns are short monosyllabic words, like "je" /ʒə/ (I), "il" /il/ (he), "elle" /ɛl/ (she). In this case, repetitions seem to be a manifestation of speech initiation difficulty and may be considered as false starts. They were counted separately, though, because of the definition we used; only partially repeated words were counted as false starts. Had they been clustered together, we would not have found a differential pattern of impairment in L1 and L2 for repetitions nor false starts.

LJ produced more filled pauses in L1 than in L2. Pauses are considered to be indicative of cognitive or linguistic processing difficulties (Krivokapi, 2007; Davis and Maclagan, 2009). In PPA, pauses have been associated with discourse, syntactic and motor speech planning, as well as word retrieval difficulties (Wilson et al., 2010; Mack et al., 2015). Given the fact that the underlying conceptualization process is the same in both languages, this finding cannot be attributed to different level of discourse processing abilities in L1 and L2. Results from the MAIN support a similar pattern of structural discourse deficits in both languages. Similarly, it cannot be attributed to differences in motor speech planning or articulation difficulties. In fact, distortions, which have been linked to apraxia of speech (Ogar et al., 2007; Duffy, 2013), were present to the same extent in both languages. The higher proportion of filled pauses in L1 could suggest a greater word finding problem in L1 compared to L2. However, LJ produced more nouns (as a proportion of narrative words) in L1 than in L2, while proportion of verbs was the same in L1 and L2. Furthermore, LJ used words of higher frequency in L2. This may indicate different levels of proficiency in L1 and L2. It must be noted here that lexical diversity was similar in both languages and that LJ made more semantic errors in L2. Greater number of filled pauses in L1 than in L2 may thus be explained with respect to the use of low frequency words and complex syntactic structures (Levelt, 1983; Ferreira et al., 1996), which is the case for the L1 narratives.

LJ produced a higher proportion of closed class words in L2 than in his L1 narratives. Nevertheless, this result must be interpreted by taking into account the increased rate of pronouns in L2. The proportion of pronouns was almost double in L2, but this can be explained by the underlying differences between French and Greek. As previously mentioned, Greek is a null subject language, whereas in French the inclusion of a subject is obligatory, and pronouns are commonly used to denote the subject in a sentence. Moreover, in the story retell task in L2, LJ was repeatedly using a double subject (both a noun and a pronoun as a subject), e.g., "The boy he was...," "the mouse it went...," The frequent use of subject doubling (double subject marking) may have inflated this measure.

In terms of discourse productivity, LJ produced longer narratives in L2 than in L1. However, proportion of narrative to total words was higher in L1 than in L2. This suggests that he was more efficient in getting his message across in L1 than in L2. Grammatical productivity was also better in L1. His sentences in Greek were more elaborate and complex, as indicated by the higher elaboration and embedding indexes in L1.

GENERAL DISCUSSION

Summarizing the information in respect to language acquisition and use, LJ is a late bilingual speaker who acquired French in adulthood through formal instruction and a 7-year-long day-to-day exposure in a French language environment. He has been using both Greek and French on a daily basis ever since, residing in a Greek-speaking country. Taking into account his wife's evaluation of level of proficiency in L1 and L2, and current exposure to both languages, Greek, LJ's first language, is his dominant language. Greek was designated as his more proficient language on the global measure of language proficiency and received a higher total score on task specific measures (11/30 in comparison to 10/30 for French). LJ has never attained fluency in reading and does not write in French. However, LJ was evaluated as being equally proficient in speaking in both languages. Language exposure to the two languages was rated as equal on the respective global measure, whilst, across different settings, language exposure to Greek (28/60) was higher than to French (21/60). Yet, the same extent of exposure to L1 and L2 was reported for interaction with his family. Even though there are skills in which LJ is equally competent in both languages and settings in which both languages are used at the same extent, taken together these results suggest that Greek is his dominant language. These results underly the complexity of the bilingual experience and illustrate the difficulty in determining language dominance that has been attested by several researchers (Treffers-Daller, 2015).

In the present study, we predicted a similar pattern of impairment in both languages and a greater impairment in L2. Altogether, results suggest a slightly worse performance in LJ's second, non-dominant language for lexical and grammatical production and the presence of a similar pattern of impairment in both languages. Our predictions are therefore only partially supported.

According to Ullman (2001), L1 lexical processing is based on declarative memory, whereas syntactic and morphological processing on procedural memory. This is also the case for L2 when it is acquired at an early age. Given the fact that LJ is a late bilingual speaker, we would expect him to rely more on declarative memory for complex syntactic and morphological processing in L2 and on procedural memory processes for grammatical processing in L1. Increasing reliance on explicit processing for L2 could also be expected because French was learned formally (Paradis, 1994). Ullman (2001) has proposed that with extended practice and higher proficiency, L2 grammatical processing may increasingly rely on procedural memory.

However, a similar pattern of performance in L1 and L2 indicates that the same organizational principles underlie the two languages (Filley et al., 2006; Hernández et al., 2008; Druks and Weekes, 2013). In a late bilingual person with different levels of proficiency in L1 and L2, like LJ, similar patterns of impairment in both languages seem to indicate shared neural representations for the two languages. This conclusion is in line with the convergence hypothesis (Abutalebi and Green, 2007; Abutalebi, 2008) which posits that L1 and L2 depend on the same neural mechanisms and that L2 lexical and grammatical representations converge to L1 representations.

This model also predicts differences between L1 and L2, as late bilingual speakers need to recruit additional cognitive control resources to process their L2. Under this theoretical account, increased processing demands exist for LJ because French is his non-dominant language. Differences between L1 and L2 may also be attributed to impaired control processes due to the underlying pathology of the nfvPPA. The executive deficit reported on neuropsychological assessment may account for the differences between the two languages. The cross-switching errors which were evident in the L2 personal narrative task support impairment in control functions. Cognitive control of L2 processing has been associated with the prefrontal cortex, the anterior cingulate cortex and the basal ganglia (Abutalebi and Green, 2007). Atrophy in the nfvPPA extends with disease progression into these regions, prefrontal cortex and anterior cingulate regions in particular (Grossman, 2010; Mesulam et al., 2014).

The fact that no significant differences were found between L1 and L2 seems to contradict our hypothesis. It must be noted however that long exposure to L2 and daily use of L2 at work and home may have played a role in preserving discourse abilities in L2. LJ uses and is exposed to French now for 36 years. Such a degree of exposure and use may play a determining role in L2 preservation. In fact, Abutalebi et al. (2015) found that differences between L1 and L2 suggesting an age of L2 acquisition effect are not present in elderly individuals. Nanchen et al. (2017) examining preservation of L1 and L2 in an immigrant population of late bilingual speakers with dementia, found that languages were equally preserved. They concluded that for elderly individuals, exposure and immersion are the main determinants of language preservation.

Our findings are consistent with a previous report (Zanini et al., 2011) of an early bilingual speaker with nfvPPA, where a decline in connected speech was found in both languages (Friulian and Italian), with the second language being impaired to a greater, but not to a significant degree. A qualitative similar pattern of deficits in L1 and L2 has been reported by Hernández et al. (2008) in an early, highly proficient Spanish-Catalan bilingual speaker with nfvPPA and Filley et al. (2006) in an early, proficient Chinese-English bilingual person with lvPPA. The only study which has investigated language abilities in a late bilingual speaker with nfvPPA was the study by Druks and Weekes (2013). Although grammatical production was not assessed, a parallel deterioration of lexical retrieval and grammatical knowledge in L1 (Hungarian) and L2 (English) was reported. This finding across two languages from different language families (Uralic and Indo-European, respectively) is similar to ours in that LJ was impaired, compared to controls, on both lexical and grammatical measures in his native language (Greek) and a parallel pattern of impairment was found in L2 (French), two structurally different languages albeit within the same family of languages.

In conclusion, we have found that LJ was impaired in lexical, discourse and grammatical productivity measures in his native language, Greek. A similar pattern of impairment was evident in his second language, French. Both L1 and L2 were affected to a similar degree. Lengthy exposure to L2 and regular activation of L2 through daily use may explain the preservation of discourse abilities in this non-dominant language. Connected speech analysis using QPA, fluency variables and error analysis has enabled the documentation of speech and language deficits present in this case of the nfvPPA and the comparison of performance between the participant's languages.

A growing body of literature indicates that behavioral interventions in PPA can result in improvement of the targeted language function, although there are generalization and maintenance issues (Cadório et al., 2017). Research on bilingual aphasia rehabilitation after stroke has yielded inconsistent results regarding the pattern of cross-linguistic therapy effects (Goral and Conner, 2013). Evidence suggests that cross-language transfer of treatment gains is easier between two highly proficient languages, and from a less-proficient language to a more-proficient language (Ansaldo and Saidi, 2014). However, cross-language transfer also depends on factors such as postmorbid proficiency levels and linguistic similarity between languages (Goral et al., 2012). These data underline the clinical importance of determining language dominance and performance in both languages in bilinguals with PPA.

One limitation of the present study is the size of the speech samples. A minimum of 150 words has been suggested for QPA (Berndt et al., 2000). However, it was difficult to obtain samples of this size without extensive prompting. A second methodological limitation was the lack of control subjects. Ideally, neurotypical Greek-French bilingual individuals should have served as controls for this study. Furthermore, performance was assessed at one time point for both languages. Although we have data that show cognitive decline, we have not evaluated language performance at two time points. Thus, no conclusions can be drawn about the pattern of decline in each language and across languages. Finally, a factor that may have influenced results in L2 is the fact that LJ was assessed in both languages by the same Greek-speaking clinician proficient in French. We know that healthy bilingual speakers' language choice is influenced by the social context and the linguistic background of the interlocutor (Blanco-Elorrieta and Pylkkänen, 2017). Nevertheless, code-switching was observed only during the personal narrative in French. It could be a task related effect explained by LJ's difficulty in accessing the relevant words in French when talking about his daily job routine.

This study calls attention to factors such as language dominance, proficiency, patterns of use, and exposure to a language. These factors play a key role in assessing bilingual individuals with PPA and making clinical decisions based on the underlying linguistic and cognitive features.

AUTHOR CONTRIBUTIONS

NK and MK designed the study. JP conducted the initial and follow-up assessments of the participant. Speech and language evaluation and QPA analysis was completed by NK. MK reviewed the QPA analysis. The manuscript was drafted by NK. Co-authors contributed to the final version of the manuscript.

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