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Disfluencies in Parkinson's Disease. A study on Italian early-stage patients[1]

Although Parkinsonian speech is often described as "disfluent", a detailed description of disruption phenomena in Parkinson's Disease (PD) has not been provided so far. The aim of this study is to identify uses and patterns of disfluencies in early-stage Italian PD subjects. The monological speech of 10 PD patients and 10 age- and sex-matched healthy subjects, all Italian native speakers, was annotated, distinguishing between Forward-Looking Disfluencies (silent pauses, lexical and non-verbal fillers, and prolongations, useful to plan message delivery) and Backward-Looking Disfluencies (repetitions, insertions, deletions, and substitutions, used by the speaker to edit something already uttered). PD and healthy productions were compared on four parameters: the number and frequency of disfluencies; their main functions; the syntactic positioning of the items; the duration of silent pauses, filled pauses, and prolongations. Results highlight the presence of specific uses, types, and characteristics of disfluency phenomena in Italian Parkinsonian speech, even at a very early stage.

Keywords: disfluencies, Parkinson's Disease, repairs, hesitations.

1. *Introduction*

Parkinson's Disease (PD) is the second most common movement disorder, originating in the basal ganglia and resulting from degenerative loss of dopaminergic neurons in the *substantia nigra pars compacta*. It affects more than 2-3 % of the population of the world over 65 years of age (De Lau, Breteler, 2006). Caused by poor muscular activation and coordination, hypokinetic dysarthria is one of PD early symptoms and consists of "abnormalities in the strength, speed, range, steadiness, tone or accuracy of movements required for breathing, phonatory, resonatory, articulatory or prosodic aspects of speech production" (Duffy, 2013: 4).

PD-related dysarthria includes, therefore, a range of voice modifications (increased voice nasality, increased acoustic noise, vocal tremor, harsh and breathy voice quality) and speech impairments, both at the segmental and suprasegmental level: imprecise consonantal articulation and reduced vowel space area, reduced voice intensity, significantly narrower tonal range (mono-pitch), mono-loudness,

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impaired speech rate and rhythm, longer silent pauses (see, among others, Darley, Aronson & Brown, 1969; Goberman, Coelho, 2005; Skodda, Visser & Schlegel, 2011; Fivela, Iraci, Sallustio, Grimaldi, Zmarich & Patrocinio, 2014).

Even though Parkinsonian dysarthric speech is usually defined as "disfluent", especially in its advanced stages, the literature still does not provide a clear understanding of the specific characteristics of disrupted PD speech (Goberman, Blomgren, 2003). Recent studies on different languages have focused on the occurrence of stuttering-like disfluencies (i.e., one-syllable word repetitions, sound and syllable repetitions, sound prolongations, and blocks) in mild-to-severe PD patients (Goberman, Blomgren & Metzger, 2010; Juste, Sassi, Costa & de Andrade, 2018). They were examined in different speech tasks and their productions were compared to those of healthy speakers and individuals with developmental stuttering. Results seem to support the relationship between stuttering and the functions of the basal ganglia and dopamine, reporting significantly greater disfluency percentages in PD patients than in the healthy control group, especially in the case of within-words disfluencies (stuttering occurring on part of a word) and in the monologic speech task.

A significative effect of the speech task on the quantity and quality of disfluency phenomena was also reported in a case study on a severely disfluent PD patient, native speaker of American English (Van Lancker Sidtis, Cameron & Sidtis, 2012). Disfluent speech was quantified and compared for conversation, two forms of repetition, reading, recited speech, and singing: while phonetic and syllabic dysfluencies appeared across all tasks, due to an impairment in motor planning of speech segments, lexical dysfluencies, related to linguistic planning, were found to be more frequent in spontaneous speech.

The results of another research, conducted on 10 Polish PD patients, reported no differentiation in the speech disfluency severity between different tasks, thus supporting the essentially organic background of disfluency phenomena in PD speech (Półrola, Góral-Półrola, 2015). However, a positive correlation between the frequency of disfluencies and the duration of the disease was found, as also observed for repetitive speech phenomena (both hyperfluent and dysfluent) in a study conducted on 53 PD patients by Benke, Hohenstein, Poewe & Butterworth (2000).

The effect of Levodopa medication on disfluencies has also been studied, to refute or support the so-called "excess dopamine hypothesis", according to which increased levels of dopamine should lead to the development of stuttering in PD speakers, with controversial results (Im, Adams, Abeyesekera, Pieterman, Gilmore & Jog, 2019).

More generally, early studies on speech errors and disfluencies were driven by the observation that these elements could work as predictors of cognitive impairment in pathologies, such as dementia and Alzheimer's disease (König, Satt, Sorin, Hoory, Toledo-Ronen, Derreumaux, Manera, Verhey, Aalten, Robert & David, 2015; Wang, Lian, Pan, Yan, Zhu, Ng, Wang & Yan, 2019). Being aware that typical speech also commonly includes phenomena like repetitions, pauses, and self-repairs, the first classifications of the phenomena ascribable to the category of "disfluencies" were defined trying to distinguish between disfluencies in typical and atypical speech (Johnson, 1961; Wingate, 1987). Later on, numerous studies concerning different languages have shown that disfluency phenomena are quite normal and pervasive in human spontaneous speech with frequencies of occurrence approximately averaging between 6 to 10 occurrences per 100 words (see Lickley, 2015). Moreover, it has been acknowledged that speakers may use some of these phenomena to manage and monitor their own speech production. Indeed, speakers may use silences, lengthen segmental material, or produce non-verbal or lexical fillers as a means of gaining extra time when they need it for the planning and construction of the upcoming message. Moreover, speakers may decide to edit or repair part of their already produced message through deletions, substitutions, or insertions (Allwood, Nivre & Ahlsén, 1990; Ginzburg, Fernández & Schlangen, 2014). Hence, some of the elements traditionally included in the heterogeneous class of disfluencies may actually provide valuable information on speech planning and discourse structuring and have been thus described as "speech management phenomena" (Allwood *et al.*, 1990).

Given these observations, investigating differences between the patterns of disfluency phenomena in PD and in typical speech may provide a privileged perspective for distinguishing and analysing uses of disfluencies phenomena progressively acquired with the outbreak of the disease and associated with cognitive, linguistic, and motor deficits resulting from the damages in the central nervous system.

In this framework, the present study aims at providing preliminary observations on the specific characteristics of speech disfluency phenomena produced by Italian subjects with early-stage PD. The starting hypotheses in this regard are essentially two: that monological spoken productions of early-stage PD patients and those of healthy individuals may differ with respect to disfluency phenomena; that this difference does not have to do with frequency but with functional, positional and/or durational aspects of disfluent speech.

2. *Method*

2.1 Participants

The data for the present research were collected from 20 Italian native speakers residing in the Campania region: 10 participants with idiopathic non-demented PD were involved (5 males, 5 females; mean age= 65), along with 10 age-matched Healthy Controls (HC, 6 males, 4 females; mean age= 67).

The patients were recruited at the Movement Disorders Unit of the First Division of Neurology at the University of Campania "Luigi Vanvitelli", from a cohort of subjects with no history of previous language and speech disorder. The diagnosis of PD was based on the modified diagnostic criteria of the UK Parkinson's Disease Society Brain Bank (Gibb, Lees, 1988). Inclusion criteria were as follows:

(1) PD onset after the age of 40 years, to exclude early-onset parkinsonism; (2) a modified Hoehn and Yahr (mH&Y) stage ≤ 2.5; and (3) disease duration ≤ 4 years. Exclusion criteria were as follows: (1) relevant cognitive impairment associated with PD assessed with the Montreal Cognitive Assessment (MoCA, Folstein, Folstein & Mchugh, 1975); (2) major depression, minor depression, and dysthymic disorder according to DSM-V criteria, rated by means of the Beck Depression Inventory (Beck, Ward, Mendelson, Mock & Erbaugh, 1961); and (3) any other neurological disorder or clinically significant medical condition.

All patients underwent an extensive assessment with PD-related scales. The presence and the severity of motor impairments were assessed through the Unified Parkinson's Disease Rating Scale part III (UPDRS III), which includes the evaluation of clinically significant speech difficulties (item III.1). The enrolled patients presented mild to moderate motor (speech) impairments at the moment of data collection.

The demographic and clinical data of patients with PD, together with the characteristics of the HC group, are reported in Table 1.

Table 1 - *Demographic and clinical features of patients with PD and HC*

		PD (n= 10) *Mean ± SD*	*HC (n= 10)* *Mean ± SD*
Demographic data	Age	65±11	67±6
	Sex (M/F)	5/5	6/4
	Disease duration (months)	31.7±18.9	
Clinical data	mH&Y stage	1.9±0.5	
	UPDRS III	25.0±8.2	
	MoCA total	24.4±4.3	
	BDI	5.2±6.2	

H&Y: Hoehn & Yahr; UPDRS: Unified Parkinson's Disease Rating Scale; MoCA: Montreal Cognitive Assessment; BDI: Beck depression inventory

2.2 Speech data collection

All participants were asked to perform a monologic task and they were recorded on a standard personal computer in a quiet room at University "Luigi Vanvitelli," by means of the software Praat (Boersma, Weenink, 2021) at a 44,100 Hz sampling rate. PD patients and HC speakers were asked to talk about positive and negative aspects of the place where they lived at the moment of the recording. All the subjects were invited to speak in their normal, conversational voice and at comfortable loudness. Sociolinguistic information on each speaker was also obtained through a questionnaire and all subjects gave written consent to the data collection procedure.

This study concerns the analysis of a subset of approximately 22 minutes of PD and 17 minutes of healthy speech which is part of a larger corpus. The latter amounts to approximately 105 minutes speech including read and spontaneous monologic speech and was already the object of a rhythmical analysis in a previous work (Maffia, De Micco, Pettorino, Siciliano, Tessitore & De Meo, 2021), which highlighted a comparable articulation rate in the two groups of speakers. The articulation rate was calculated by measuring the VtoV parameter, i.e., the mean duration of the intervals between two consecutive vowel onsets (Pettorino, Maffia, Pellegrino, Vitale & De Meo, 2013): in the PD speech the mean VtoV value was 0.188 s, in healthy speech it was 0.185 s. Moreover, mean data on speech time composition showed no statistical difference in the percentage of disfluent time on the total utterance (PD= 14 %; HC= 11 %). Starting from these observations, the present work aims at providing a more detailed analysis of the way PD and HC speakers may use disfluency phenomena to manage their speech production.

2.3 Analysis

The objects of the analysis are speech elements that are commonly referred to as "disfluencies", though this is a highly debated umbrella term used for a wide range of phenomena. In this study, we observe elements that have been described as "speech management phenomena" as they may be used by speakers to monitor and effectively manage the online processes of speech planning, coding, articulation, and reception (Levelt, 1989). As more extensively described in (Schettino, Betz, Cutugno & Wagner, 2021; Schettino, 2022), each phenomenon is identified and annotated on three main levels[2], with the context of occurrence being discriminative for the identification and annotation processes[3]. On the first level, the disfluencies' macro-structure is labelled: the region to be repaired (*Reparandum*, RM), the repaired one (*Reparans*, RS), and the one where the delay occurs (*Interregnum*, IM). The second level is for the identification of specific items: *Deletions* (DEL), *Insertions* (INS), *Substitutions* (SUB), *Repetitions* (REP), *Silent Pauses* (SP), *Prolongations* (PRL), *Filled Pauses* (FP), and *Lexicalized Filled Pauses* (LFP). On the third level, each item is assigned its main function: *Forward-Looking* (FLD), when their occurrence suspends the speech delivery thus gaining time for speech planning processes; *Backward-Looking* (BLD), when they trace back to and alter speech strings that have been already uttered.

The analysis concerns the comparison of Parkinsonian (PD) and Health Control (HC) speech regarding the following parameters:

- the frequency of disfluent items;
- the main contextual function of the occurrences (BLD, FLD);
- the syntactic positioning of the items, i.e.,
  - within words (WTH – W), e.g., «ma io no+ <ehm> non ho i cani»,
  - within phrases (INTRA – S), e.g., «la zona dei paesi<ii> scandinavi»,

[2] The data were transcribed following the guidelines provided by Savy (2005).

[3] Accordingly, no fixed minimum duration threshold was set to detect neither silent pauses nor prolongations. Then latter were defined as silences or segmental lengthening perceived as marked in their context of occurrence. The reliability of such system was verified by measuring the agreement between two expert annotators (Cohen's k = 0.82, good agreement, Landis, Koch, 1977; in Schettino, 2022).

  - between phrases (INTER – S), e.g., «ci sono <mh> <sp> molti monumenti»,
  - between clauses (INTER – C), e.g., «ci stanno le persone <eeh> che ti conoscono»;
- the duration of Silent Pauses, Filled Pauses, and Prolongations.

The annotation was conducted using the ELAN software for multi-level linguistic annotations (Sloetjes, Wittenburg, 2008). Considering the high degree of individual variability that characterizes speech productions, the statistical significance of the results is tested by building different Generalised Linear and Linear Mixed Models (lme4, Bates, Maechler, Bolker & Walker, 2015) to observe the relevant and consistent features net of such variability. The models were built by considering the previously listed analysis parameters (frequency of occurrence, main contextual function, syntactic positioning, duration) as dependent variables. The effect of the health condition (PD or HC), the subjects' biographical data (sex and age), and clinical variables (mH&Y stage, UPDRS III, MoCA total, BDI) on the chosen parameters was tested by including them in the models as "explanatory" independent variables. Then, since the analysis is based on the occurrences of disfluency phenomena and each speaker produced definitely more than one phenomenon, the models included Speaker as a random effect to control for individual variability.

3. *Results*

3.1 General frequencies and main function

The analysis concerned 581 disfluency phenomena. In the observed data, they were slightly more frequent in the PD productions (294) than in the HC productions (281).

Also, when considering the different types of phenomena (Figure 1), the most frequent are repetitions, prolongations, and silent pauses. Comparing PD and HC speakers' productions, PD speakers only seem to produce more substitutions and repetitions, but no significant difference emerges (Est.=0.28, SE=2,71, z=0,11, p=0.91). Nonetheless, when considering the main function associated with each item (Figure 2), both PD and HC speakers produce considerably more Forward-Looking Disfluencies than Backward-Looking ones, but PD speakers are found to produce significantly more Backward-Looking Disfluencies than the speakers of the control group (Est.=0.87, SE=0.41, z=2.1, p=0.035). No significant effect is exerted by the biographical variables (sex and age). As for the clinical variables, among PD, cognitive scores significantly affect the use of BLD, the latter are significantly more with lower scores (MoCA, Est.=0.11, SE=0.04, z=2.74, p= 0.006, see Figure 3).

Figure 1 - *Frequency of occurrence of the different types of phenomena per condition and grouped by main function: Backward-Looking (left panel), Forward-Looking (right panel)*

Figure 2 - *Frequency of occurrence of Backward-Looking and Forward-Looking phenomena per condition*

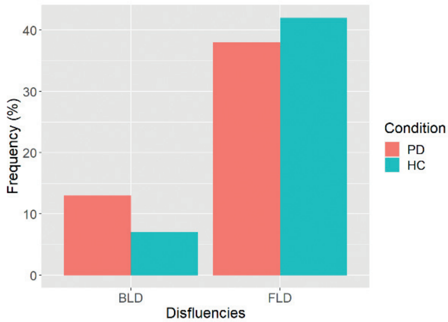
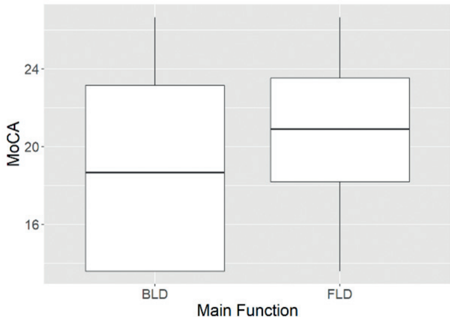


Figure 3 - *MoCA score of Backward-Looking and Forward-Looking phenomena*



3.2 Positioning

The distribution of phenomena with reference to syntactic structures (clauses, phrases, and words) also seems quite different between the PD and the HC groups (Figure 4). In this case, the statistical analysis was conducted by fitting GLMMs with the positioning considered as binomial dependent variable. The model where the outcome had one level for within-word occurrences and the other level grouped the occurrences in the other positions yielded significant results for both Condition and Main Function. Namely, disfluency phenomena occur significantly more frequently within words, that is interrupting word production when produced by PD speakers (Est.=1,99, SE=0,67, z=2,94, p=0.003) and when associated with a BLD function (Est.=1,6, SE=0,46, z=3,41, p=0.0006). Since the main function associated with disfluency phenomena was found to exert an effect on the positioning of the occurrences, we further tested BLD and FLD separately. Backward-Looking phenomena occur significantly more frequently within words and less frequently between words in PD productions than in HC speech (Est.=1,93, SE=0,71, z=2,73, p=0.006). Similarly, Forward-Looking phenomena interrupt words significantly more frequently in PD than in HC speech (Est.=2,49, SE=0,61, z=4,06, p=<.0001).

Figure 4a-d - *Position of Backward-Looking and Forward-Looking phenomena produced by PD and HC speakers*

Figure a - *PD – BLD*

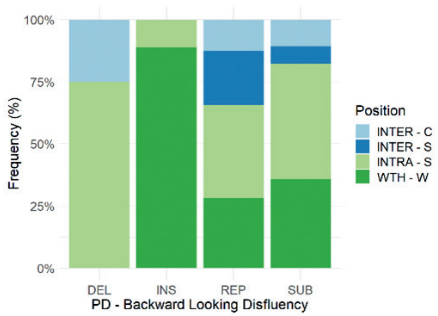


Figure b - *HC – BLD*

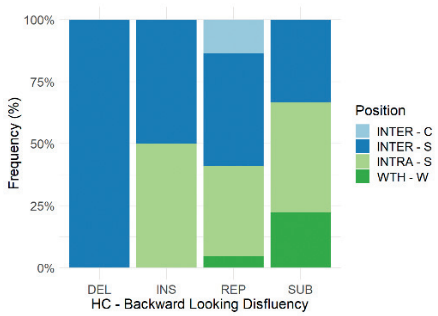


Figure c - *PD – FLD*

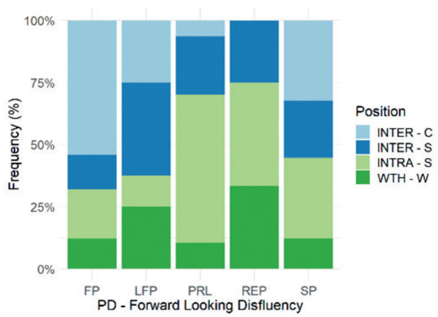
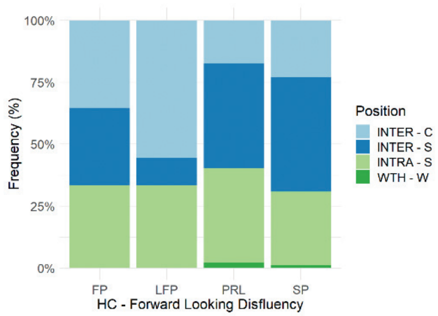
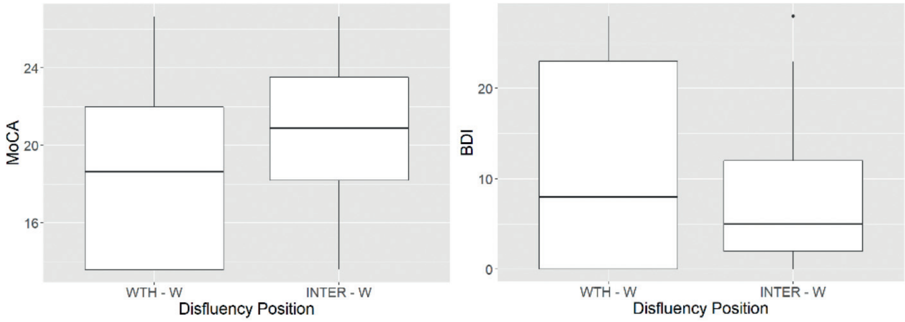


Figure d - *HC – FLD*



Considering the clinical variables among PD (Figure 5), position was found to be affected by cognitive scores (MoCA), phenomena interrupting words being significantly more frequent with lower cognitive scores and phenomena non-interrupting words being more frequent with higher scores (Est.=0.19, SE=0.06, z=3.19, p=0.001). Conversely, within-words phenomena were significantly more frequent with higher depression scores (BDI, Est.=-0.10, SE=0.04, z=-2.3, p=0.02).

Figure 5 - *MoCA score (left) and BDI score (right) per phenomena within-word or between-word position*



3.3 Duration

As reported in Table 1 and illustrated in Figure 6, on average, Forward-Looking phenomena produced by PD speakers tend to be slightly longer than those produced by HC speakers. However, when considering the different types of phenomena, a significant difference only emerged for Silent Pauses (Est=211.28, SE=78.27, t =2.699).

Figure 6 - *Duration of Silent Pauses, Prolongations and Filled Pauses per condition*

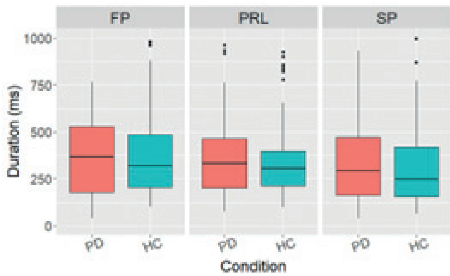


Table 2 - *FLDs duration values per condition: count, mean, standard deviation, standard error of the mean, and confidence interval (default 95 %)*

FLD	*Cond*	*n*	*Dur(ms)*	*sd*	*SE*	*ci*
SP	PD	74	521	555	64	128
SP	HC	89	343	266	28	100
PRL	PD	77	372	372	25	50
PRL	HC	92	344	344	19	38
FP	PD	50	472	360	51	102
FP	HC	50	468	352	49	100

4. *Discussion*

This work has proposed an approach to describe the characteristics of disfluency in Italian patients with early-stage PD by considering the way speakers use disfluency phenomena to manage their spontaneous speech. More specifically, we distinguished between Forward-Looking and Backward-Looking Disfluencies, the former associated with a suspension of message delivery and a need for extra time for language planning, the latter used to edit something that has already been said.

As previously hypothesized, this distinction highlighted significant differences, although the considered PD and HC speech did not differ much in the number of occurrences of speech disfluency phenomena, as also observed by Maffia *et al.* (2021), and both PD and HC speakers were found to more frequently resort to phenomena suspending the speech delivery. In fact, the patients needed to repair something already uttered significantly more frequently than healthy subjects. In particular, they more frequently produced Substitutions, to edit something already uttered and deemed inappropriate to a certain extent, and Repetitions with a bridging function (Shriberg, 1995).

Relevant differences between the production of disfluency phenomena in the PD and HC groups of speakers also concerned the syntactic positioning of disfluencies: while healthy speakers more frequently produce these phenomena between larger units such as clauses and between phrases, PD speakers interrupt words with within-word phenomena significantly more frequently, which corroborates previous findings in the literature (Juste *et al.*, 2018; Goberman *et al.*, 2010).

Moreover, PD patients were observed to produce slightly fewer Silent Pauses and Prolongations, but significantly longer pauses than healthy speakers, which is also in line with previous findings (Darley *et al.*, 1969; Goberman, Coelho, 2005).

At a closer inspection, some clinical variables (cognitive and depression scores mainly) were also found to play a significant role in explaining the observed tendencies in the characterization of PD speakers' production of disfluency phenomena. Although all patients scored above the MoCA threshold and below the BDI threshold that assesses the outbreak of dementia or depression respectively, the increased need for repair observed in PD patients, as compared to healthy speakers, is greater the worse the cognitive condition assessment is. Just as well, PD speakers' need to interrupt words is greater the lower cognitive and the higher depression scores are. These findings support the idea that the features observed as characteristic of PD speech can be considered as a result of a progressive process of deterioration of physiological conditions which determines the development of different strategies to manage the own speech production.

5. *Conclusions*

To conclude, the preliminary findings reported in this study have highlighted that it is the study of the way disfluency phenomena are used by speakers to manage their speech rather than mere frequency observations that may provide relevant information concerning the features of Parkinsonian speech and the conditions of patients.

It seems that, even at a very early stage of the disease, when patients' speech is still completely intelligible and the effects of dysarthria are not yet perceptible, both motor and linguistic planning and processing are somehow already altered, as the nature and the occurrence of disfluencies reveal. When compared to healthy speakers, PD patients need and spend more time repairing their own utterances, often interrupting the production of words and producing significantly longer silent pauses.

A plausible explanation has to be sought at the intersection of cognitive impairments and motor modifications in speech production, which both seem to affect communication abilities from the very beginning of Parkinson's Disease and even in the absence of dementia (Rohl, Gutierrez, Johari, Greenlee, Tjaden, Roberts, 2022). However, in light of the limited sample size, the results of this study require caution in their interpretation and need further and more detailed replications.

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