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Investigating Verbal Repetition in Persian-Speaking Patients with the Semantic Variant of Primary Progressive Aphasia Compared to Healthy Individuals

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ABSTRACT

Introduction: Language deficit is regarded as one of the most important hallmarks of primary progressive aphasia. This study aims to analyze the nature of verbal repetition ability in a group of patients suffering from the semantic variant of primary progressive aphasia and investigate their cognitive capability.

Materials and Methods: In this cross-sectional study, 12 patients of the semantic variant of primary progressive aphasia and 12 healthy individuals accomplished a set of tasks, including naming, word comprehension, digit span, repetition of multisyllabic single words, monosyllabic word span under similar or dissimilar phonological conditions, and sentence repetition.

Results: The patients exhibited some degree of impairment with diverse patterns in each task, although the accomplishment of the sentence repetition task was the most challenging activity for the primary progressive aphasic patients of the semantic variant. However, the healthy individuals performed well in all tasks.

Conclusion: The recognition of the quantity and quality of the cognitive deficit in the semantic variant of primary progressive aphasics can illuminate the nature of this disease and contribute to a better diagnosis of this disease. Furthermore, it can assist speech therapists and neuropsychologist to recruit appropriate therapies and employ better techniques for the diagnosis and cure of these patients.

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1. Introduction

As a multifaceted neurodegenerative syndrome, primary progressive aphasia (PPA) is a type of non-Alzheimer dementia, characterized by gradual deterioration of language performance [1]. The semantic variant of primary progressive aphasia (sv-PPA) has a unique linguistic pattern. In this type of PPA, semantic manifestations are gradually deteriorated, even though speech output is preserved. The patient demonstrates a normal speed of speech with few syntactic errors; however, they generate an abundant number of closed-class items, such as pronouns, verbs, and nouns while suffering from lexical retrieval deficit [2, 3]. In this type of aphasia, which is also known as semantic dementia, frontotemporal lobar degeneration TDP-43 pathology is a commonly affected area. Furthermore, major atrophy of the left hemisphere, deterioration of the perisylvian language network in the superior and middle temporal gyri, and the atrophy of anterior components in the face and object recognition network in the inferior and medial temporal lobes are considered the main pathological characteristics of these patients [4, 5].

One of the most important challenges that clinicians have in the diagnosis of this syndrome is its idiosyncratic nature; that is, in some circumstances, patients afflicted with this syndrome demonstrate minor or overlapping characteristics with the other two types of sv-PPA, which makes their classification different [6-8]. Because of the above-mentioned challenges, the special data that we could obtain through the conduction of verbal repetition tasks was regarded as a complementary tool for the prevalent diagnosis of the disease [7]. In addition, the achieved results could elucidate what kind of cognitive deficit would impede these patients to perform such cognitive and linguistic tasks accurately.

In the verbal repetition tasks, meaningful segments, including words, numbers, phrases, or sentences are auditory presented to the participants. Then, the participants are required to immediately recall and verbally utter these segments. The complexity of these tasks might vary according to the parameters, such as length (whether the participants are required to repeat single words or multiple words, phrases, or sentences). Also, the phonological characteristics of the words might play a determining role in task complexity. As a result, the accomplishment of each specific type of verbal repetition task would entail the integration and association of diverse cognitive mechanisms [9]. In other words, single-word repetition requires phonemic integration as well as motor execution of a coordinated articulatory plan [10, 11]. The correct administration of word or number span tasks requires cognitive overload controlled mainly by short-term memory [12]. Also, the phonological similarity effect can impact the accurate performance of participants in word span tasks as previous literature demonstrates the advantage of phonologically dissimilar elements over phonologically similar ones [13-15]. However, among all the above-mentioned tasks, sentence repetition is the most challenging and complex item because the satisfactory execution of this task requires the accurate manipulation of the mentioned factors in the previous tasks and it entails the specific cognitive capacity to combine small segments into meaningful chunks [16, 17]. In this task, which has been designed to measure the major linguistic capability of individuals, the participants are expected to repeat different types of sentences with diverse lengths and complexity [18].

In this study, through the conduction of different types of tasks, including digit span, word spans, repetition of multisyllabic words, and sentence repetition, we attempted to scrutinize diverse cognitive components involved in the sv-PPA. Our specific objective in this study, considering different morphological and syllabic characteristics of the Persian language compared to other Indo-European languages, was to compare the verbal repetition capability of sv-PPA in the sentential and word level with varying complexity. We predicted that the patients afflicted with sv-PPA should demonstrate normal performance in all tasks of verbal repetition because their speech production capacity is intact; however, as semantic representations are disrupted in these groups of patients and their working memory capacity diminishes, they might be incapable of establishing meaningful relationships among different meaningful units or chunks [9, 19]. Accordingly, as the chunking process by which linguistic segments are combined and concatenated is disrupted in these patients, the repetition of meaningful sentences compared to the repetition of separate words might be more challenging for them.

2. Materials and Methods

Study participants

This is a cross-sectional descriptive-analytical study. The method for selecting the study participants was the convenient sampling method, based on which 12 patients visiting Iran Dementia and Alzheimer's Association from April to September 2019, along with 12 education-and age-matched healthy individuals were selected. All the patients were categorized as PPA based on the

results of diverse neuropsychological tests, their magnetic resonance imaging (MRI), and neuropsychological assessments. Accordingly, at the initial stage, their disease history was surveyed and a physical examination was conducted by a neurologist. Then, the patients underwent MRI whose results culminated in the diagnosis of the disease. The criteria for the diagnosis of sv-PPA were based on the current international consensus recommendation [20]. Also, the patients whose scores were below 50 on the bedside version of the Persian aphasia battery (P-WAB-1) [22] were selected. Other inclusion criteria were as follows: 1) The lack of abnormalities in their MRI; 2) The lack of addiction to alcoholism or drug abuse; 3) Less than 4 years since the onset of PPA; 4) The lack of severe cognitive and psychological disorders; 5) Being right-handed; 6) The pathology of all patients corroborated frontotemporal lobar degeneration and also the deterioration of the perisylvian language network in the superior and middle temporal gyri; 7) sv-PPA diagnosed by a neurologist who was expert in the area of neurodegenerative diseases; 8) Endowed with a rather intact single-word comprehension or confrontation naming capability as the decisive clinical properties of the patients. Our exclusion criteria were as follows: 1) Participants with limited Persian proficiency; 2) Patients with advanced disease who were incapable of repeating even a pair of words; 3) Patients with abnormalities on their MRI, suffering from traumatic brain injury, stroke, visual, or auditory deficits; 4) Affliction with other neuropsychological diseases, such as depression, anxiety, or obsessive-compulsive behavior. Ultimately, having consulted with the neurologists and reviewed the neuropathological profile of the patients, we selected 12 patients with (sv-PPA) of semantic variety whose pathology vividly corroborated their affliction with the syndrome. Our selected subjects were all with a mean age of 67.8 years and a mean education of 13.7 years. Also, 12 age- and education-matched healthy individuals were selected as the control group.

Study instruments

Neuropsychological and linguistic tasks

The standardization procedures were conducted via performing a pilot study on 12 age- and education-matched individuals. Two autonomous neuropsychologist expressed their judgments regarding the internal consistency of the tests. The results of the Cronbach α for the forward and backward digit span tasks were 0.93 and 0.89 for each task, respectively. In the word span task under phonologically similar or dissimilar conditions, reliability of 0.96 and 0.87 were reported for each condition. Ultimately, in the sentence repetition task, the internal consistency was 0.95. Hence, the results corroborated the high reliability and suitability of our tests.

Global cognitive abilities

We recruited the Persian version of the mini-mental state examination [23] to evaluate our students' global cognitive abilities. This test evaluates the cognitive abilities of individuals, including memory, language, executive function, visuospatial capability, and working memory. The total score is 30 where higher scores demonstrate better cognitive ability of the participant. Meanwhile, the cutoff score is 23.

Single word tasks

We chose 3 tasks from the Persian aphasia battery test [24], including naming, word comprehension, and repetition of multisyllabic words. Our rationale for the selection of the two aforementioned linguistic sub-tasks was that they have internationally been regarded as the two core clinical features of sv-PPA [20]. Furthermore, their usefulness to track language decline over time in sv-PPA has also been corroborated [25]. This battery demonstrated convergent validity with the Sydney language battery, along with a reliability of 0.93 divulging its efficacy to monitor linguistic decline in the sv-PPA [25]. In the confrontation naming task, the participants were asked to name 20 diverse color photographs from different categories with graded difficulty one at a time. We scored the items that were correctly named. In the single-word comprehension task, the participants were asked to point to the picture which best matched the word uttered by the researcher. The presented pictures included 6 items that were either visually or semantically related to the target word or matched with one of 30 items in our initial naming task.

Our rationale for the execution of the repetition task was 2-fold. First, the accurate execution of the task requires the integration of diverse cognitive elements, namely, the accurate concatenation of sounds composing the words, besides the accurate function of a coordinated articulatory plan. Consequently, via the conduction of this task, the disruption or integration of diverse cognitive elements could be examined. Second, via the selection of multisyllabic words, we attempted to scrutinize whether word complexity could have a negative impact on the participants' performance [10].

In the word repetition task, the participants were expected to listen and repeat 20 multisyllabic words corresponding to the same items in the naming task (see the lists of words in Table 1). In this task, different types of phonological errors, including deletion, addition, or permutation (the displacement of neighboring phonemes) were considered. Furthermore, the participants' pauses or their frequent attempts to initiate repetition were envisaged as phonological errors.

For statistical stability, all the scores in the verbal repetition tasks were transformed into percentages. As a result, considering 20 as the maximum raw score, we considered the percentage of correct linguistic items in each task for our data analysis.

Digit span tasks

Any reduction in the capacity of working memory would lead to the individuals' malfunction in the verbal repetition tasks [26]. We recruited verbal/auditory digit span tasks from the Wechsler adult intelligence scale, third edition [27] to evaluate the working memory capacity of the participants. In the forward condition of digit span tasks, the participants were expected to recite the exact sequence of numbers uttered by the researcher. However, in the backward version of the task, the participants were required to recite the numbers in their reversed order. We derived the percentage of correct scores from the raw scores, while 16 was the maximum raw score for the forward condition and 14 was the maximum number for the backward condition.

Word span tasks

Another recruited working memory assessment tool was the word span task. Accordingly, the participants were required to recall lists of monosyllabic words presented under phonologically similar or dissimilar conditions [9]. The length of these lists increased from 2 to 4 words. Each 10-word set was equally divided into 5 lists of phonologically similar and 5 lists of phonologically dissimilar words (in case the participant could repeat the words in their exact serial position and made no phonological errors of any kind, their produced list was regarded as correct). For instance, if the list of the original words which the participant should have produced was ("del," "man," "yar," "gol"), but instead she produced ("del," "yar," "man," "gol"), although they were capable of producing all words in the set, in our scoring system, we regarded it as incorrect because they were not capable of uttering the words in their exact initial serial order. The percentage of corrected produced words for each length and phonological consideration was estimated.

Sentence repetition task

In this task, which was recruited from the Persian aphasia battery test [24], the participants were required to repeat 20 sentences of diverse lengths, varying from 3 to 24 syllables. Self-corrections or self-attempts by the participants were permitted. Again, the percentage of correctly produced sentences was estimated.

Statistical analysis

Our statistical analysis was conducted via the SPSS software, version 20. We utilized the X^2 test to evaluate the gender ratio between groups. Furthermore, concerning continuous variables, we utilized the one-way analysis of variance followed by the Bonferroni corrections to demonstrate between-group differences. Also, in the single-word tasks, to eliminate ceiling and floor effects, the Kruskal-Wallis tests, followed by the post hoc pairwise comparisons concomitant with the Bonferroni corrections at $P<0.05$ were administered.

To compare performances between the 2 groups in digit span conditions as well as word span tasks, 2 separate repeated-measures analyses of variance were conducted. The Mauchly test of non-significance was presumed as sphericity and if this postulation was violated, we used the Greenhouse-Geisser method to adjust the degree of freedom. All observed significant differences in our model were investigated utilizing the post hoc pair-wise comparisons which were adjusted with the Bonferroni corrections.

3. Results

Global cognitive abilities

The results of the mini-mental state examination demonstrated significantly lower performance of the patients (47.9±12.1) compared to the healthy individuals (92.0±2.8, $P<0.05$).

Single-word tasks

Although both groups of healthy individuals and the sv-PPA aphasia performed approximately at the same level in the repetition of single words, the experimental group performed poorly compared to healthy individuals in both tasks of word naming and word comprehension tasks ($P<0.005$).

Table 1. List of words in the word repetition task

No.	Name of Category	Word
1	Flower	Banafshe
2	Animal	Shotormorq
3	Food	Qormesabzi
4	Liquid	Shirkakau
5	Vegetables	Kalamborokli
6	Pottage	Ashesshole qalamkar
7	Color	Banafshe Arqavani
8	Color	Zard Kahrobaee
9	Meat	Kababbakhtaiari
10	Furniture	Miznaharkhori
11	Trees	Sarvekuhi
12	Fruits	Porteqallobnani
13	Cloth	Jurabshalvari
14	Cake	Mafineshokolati
15	Sweet	Keikshokolatie khis
16	State	Ashoftehal
17	State	Heiratzade
18	Building	Mehmansarayelukse jahangardi
19	Manner	Kharamankharaman
20	Ocean	Oqyanusemonjamedeshomali

Word span tasks

The overall performance of both groups deteriorated across 2 different conditions of phonologically similar or phonologically dissimilar word conditions. This malfunction in the performance of both groups also collapsed as the length of our lists increased ($F_{(10.8, 1270)}$=2424, P<0.001). This decline was statistically different across groups. However, the post hoc pair-wise comparison indicated that although the two groups performed approximately equally in the first set composed of two words, the group of the semantic variant of primary progressive aphasia performed significantly at a lower level compared to the control group when faced with the word span of 3. At the span length of 4, the experimental group demonstrated remarkably abnormal performance.

All participants demonstrated better performance on the repetition of dissimilar words than on a similar set of words ($F_{(1, 71)}$=101.4, P<0.001). However, the interaction effect between the two groups and the phonological condition did not reach statistical significance ($F_{(4, 71)}$=2.0, P=0.11). This result demonstrated that both groups had rather better performance in the repetition of dissimilar words span in the repetition of similar words. According to the length of the lists of words involved, the effect of phonological similarity was different ($F_{(2, 1.8)}$=44.6, P<0.001). Although in the span length of 2, this effect was not observed, in the other set of words, we observed a clear similarity effect. Our further analysis concerning between-group interaction, lists length, and phonological effect corroborated a statistical inclination ($F_{(8, 7.3)}$=2.0, P=0.05). The post hoc analyses demonstrated that in the phonologically similar conditions, as soon as the length

of the word lists increased in the 2 groups, the performance declined. However, this decline in dissimilar conditions occurred only between the span length of 3 and 4.

Sentence repetition task

The results demonstrated that our patients showed lower performance compared to the control group in the sentence repetition task ($P<0.005$).

Overall performance of study groups on repetition tasks

As our results showed, the patients afflicted with sv-PPA performed at above-chance level approximately on all tasks, except for the sentence repetition task in which they demonstrated severe impairment. However, the control group demonstrated rather intact performance in all tasks.

4. Discussion

The group pattern of performance in all tasks corroborated their rather intact performance in most verbal repetition tasks. Notwithstanding, our patients had a remarkably below-chance performance on the sentence repetition task. This finding is not novel given that the repetition of a sentence required lexico-semantic adjunction at the word level and it entails the amalgamation of separate lexemes observing grammatical constraints permitted in the Persian language.

Concerning single-word tasks, our patients demonstrated below-chance performance on single-word comprehension while exhibiting above-chance performance on single-word repetition, representing dominant characteristics of severe anomia. This finding reflected that their semantic manifestations were integrated [28-30].

In both forward and backward digit span tasks, the experimental group demonstrated poor performance, although, in the backward condition, they performed more poorly. These findings were not surprising given that the accurate administration of these tasks requires the associations of cognitive, linguistic, and articulatory components. Considering that our patients had more challenges in the administration of the backward digit span task compared to the forward digit span task, this could be justified because the accurate performance of the former requires the accurate function of working memory to store information tentatively and it entails the reversed manipulation of items performed by the executive function system which is impaired in these patients. These findings were in line with most recent studies [19, 31-33].

In the word span tasks, the performance of the participants was affected by the list length and phonological effect. While in the list length of 2, both groups had rather intact performance in either phonologically similar or phonologically dissimilar word spans, the experimental group demonstrated a remarkably weak performance in the other list length. As the length of word lists increased, the participants demonstrated a weaker performance. The difficulty was more clear in the experimental group, especially when they had to repeat phonologically similar lists composed of 3 or 4 words. These findings corroborated the previous research, emphasizing the determining role of the parameters of length and phonological similarity upon the performance of primary progressive aphasics of semantic variety. As our results showed, our healthy participants demonstrated poor performance on repetition tasks, committing some phonological errors [9, 25].

As mentioned earlier, concerning sentence repetition tasks, the patients performed weak performance because even though their verbal short-term memory remained intact, their semantic representations were disrupted refraining them to create and maintain a plausible meaningful relationship between words in a sentence.

5. Conclusion

As our results showed, in the word span tasks, where there was phonemic similarity among words, sv-PPA demonstrated several challenges while they had a rather intact performance on the repetition of phonologically dissimilar words. However, they performed more poorly in the sentence repetition task than in the word task because binding small segments into bigger meaningful chunks entailed complex cognitive capacity of which these patients were bereft. Henceforth, given our findings, the conduction of different span tasks as well as sentence repetition tasks (with diverse syntactic complexity) are fruitful tools with which cognitive and linguistic capacities of sv-PPA could be evaluated, and in doing so, appropriate cure for these patients could be implemented.

Study limitations

Although our results corroborated the poor performance of the sv-PPA, some caveats should be considered. First, as our sample size was small, it might not be plausible to generalize the findings to a wider context. Second, given the outstanding typological differences among languages of the world, even the conduction of the same research with the same methodology in other

languages might culminate in different conclusions. Third, if the number of syllables in the stimuli used in the word span tasks increased, i.e. polysyllabic words were utilized or if the lists of words to be repeated increased, all these changes might have different repercussions, affecting the results of the research. Fourth, had we utilized a different methodology or employed online tasks, we might have been confronted with different outcomes.

Ethical Considerations

Compliance with ethical guidelines

This study was approved by the Ethics Committee of University of Gonabad (Code: 2021/52200/1410/98). Before the conduction of the research, an informed written consent form was taken from our patients, demonstrating their voluntary participation in the study.

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Conflict of interest

The author declared no conflict of interest.

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