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ARTICLE

Speech disorders did not correlate with age at onset of Parkinson's disease

Distúrbios da fala não se correlacionam com a idade de início da doença de Parkinson

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ABSTRACT

Speech disorders are common manifestations of Parkinson´s disease. **Objective:** To compare speech articulation in patients according to age at onset of the disease. **Methods:** Fifty patients was divided into two groups: Group I consisted of 30 patients with age at onset between 40 and 55 years; Group II consisted of 20 patients with age at onset after 65 years. All patients were evaluated based on the Unified Parkinson's Disease Rating Scale scores, Hoehn and Yahr scale and speech evaluation by perceptual and acoustical analysis. **Results:** There was no statistically significant difference between the two groups regarding neurological involvement and speech characteristics. Correlation analysis indicated differences in speech articulation in relation to staging and axial scores of rigidity and bradykinesia for middle and late-onset. **Conclusions:** Impairment of speech articulation did not correlate with age at onset of disease, but was positively related with disease duration and higher scores in both groups.

Keywords: Parkinson´s disease, speech articulation, dysarthria.

RESUMO

Distúrbios da fala são comuns da doença de Parkinson. **Objetivo:** Comparar a articulação da fala de acordo com a idade de início da doença. **Métodos:** Cinquenta pacientes foram divididos em dois grupos: Grupo I consistiu de 30 pacientes com idade de início entre 40 e 55 anos; Grupo II foi composto por 20 pacientes com idade de início após os 65 anos. Todos foram avaliados pela *Unified Parkinson's Disease Rating Scale*, Hoehn and Yahr e análise perceptual e acústica da fala. **Resultados:** Não houve diferença estatística significativa entre os dois grupos quanto às características neurológicas e de fala. Análises de correlação indicaram diferença estatística significativa entre articulação da fala, estágio da doença e escores axiais, de rigidez e bradicinesia. **Conclusões:** A articulação da fala não se correlacionou com a idade de início da doença, mas foi positivamente relacionada à duração da doença e aos escores mais elevados nos dois grupos.

Palavras-chave: doença de Parkinson, articulação da fala, disartria.

Parkinson's disease (PD) is a neurodegenerative disorder characterized by loss of dopamine-producing cells and affects mainly individuals over 60 years but a young onset subtype is also well recognized[1]. Motor limitations observed in PD are often attributed to basal ganglia dysfunction associated with decreased dopaminergic input to the sensorimotor region of the striatum[2].

The progressive neuronal loss is associated with a variety of motor and non-motor deficits in PD patients. In addition to the predominant symptoms such as muscular rigidity, tremor, bradykinesia and postural instability, many patients develop a distinctive alteration of voice and speech characterized as hypokinetic dysarthria[3]. Nearly 90% of people with PD will develop voice and speech disorders during the course of the disease[4], which can have a negative impact on functional communication and result in poor quality of life[5].

Dysarthria occurs as a result of disturbances in planning and execution of speech and involves several neural mechanisms related to basal ganglia, cerebellum, supplementary motor area and frontal circuits[6]. Voice and speech disorders associated with PD are most commonly characterized by one or a combination of the following perceptual characteristics; reduced vocal loudness[7], breathy or hoarse/harsh voice quality[8], reduced voice pitch inflections or monotone voice[9], and speech impairment articulation[10]. Acoustic measurements revealed that PD patients produce undershooting of articulatory gestures[11], which leads to imprecise articulation of consonants and vowels[12]. According to some authors, these changes may be present even before the onset of more overt PD manifestations[13].

Symptoms of PD begin characteristically above the age of 50 years with a mean age of onset around 60 years but some

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patients develop PD at a younger age. Young adults confront problems that are fundamentally different from those faced by people in their sixties or seventies, and the impact of PD in early-onset patients is therefore likely to differ from that in older patients[14].

Age of onset of clinical symptoms in PD is one of several criteria employed for disease classification[15]. Age cut-offs for early versus late onset subtypes vary but 40 or 50 years of age is used as a defining threshold[16].

Although there is some evidence that age at onset may influence characteristics and progression of PD[17], differences between early and late-onset PD have not been fully explored. Some authors have suggested that late-onset PD is associated with more rapid progression, whilst a slower and more benign course should be expected in early-onset patients[18].

Although speech articulation disturbances have been considered a symptom of disease progression[19], little is known about its emergence in relation to disease stage and to global motor function. Studies on the influence of age in the clinical manifestations of PD, including speech, should contribute to a better understanding of specific symptoms and to provide better diagnostic, therapeutic and rehabilitation strategies.

The aim of this study is to compare speech articulation between two groups of PD patients according to age of onset and to correlate the degree of speech impairment with motor symptoms and disease severity.

METHOD

Patients

Fifty patients with diagnosis of idiopathic PD and preserved speech intelligibility, with or without speech complaints were included. Diagnosis was made according to the United Kingdom Parkinson's Disease Society Brain Bank Criteria[20]. Patients were classified and divided into two groups according to age at onset: group I (onset between 40 and 55 years) and group II (onset ≥ 65 years). These categories were termed middle-age onset and old-age groups, respectively. Both groups were matched for duration of disease.

Exclusion criteria included diagnosis of dementia (DSM IV)[21] or depression (UPDRS-I)[22], any surgical procedure to treat PD, hearing or language disorders and current speech treatment. Patients were asked to complete the assessment interview with the aid of caregivers.

Procedures

All participants were receiving regular dopaminergic therapy and were evaluated during the "on" phase.

Neurologic evaluation

Patients underwent a neurological examination, according to Hoehn and Yahr Scale (HY)[23] and Unified Parkinson´s Disease Rating Scale (UPDRS) part III[22]. Motor scale was divided into four domains: tremor, rigidity, bradykinesia and axial impairment. Tremor evaluation was based on item 20 and 21; rigidity was based on item 22; bradykinesia was based on items 23, 24, 25, 26 and 31; and axial score was based on items 18,19, 27, 28, 29 and 30. Motor examination was performed by a movement disorder specialist.

Perceptual analysis

Patients were asked to emit the sustained vowel /a/, to count numbers from one to 20 and were induced to comment on their speech difficulties (spontaneous speech). Samples were registered by one of the authors (AED) in a Sony™ MiniDisc, model MZ-R700 from Shure microphone, model SM-58, at a distance of 15 cm and inclined at a 45 degree angle in relation to patient's mouth. Samples were further analyzed by three assigned speech therapists (judges) considered to have expertise in PD. In order to minimize variability among evaluations, each judge was oriented to pause between analyses to avoid tiredness. Evaluations were always performed early in the morning at approximately the same time, blinded in relation to patient identification and at a threshold of 70 dB SPL. No communication among judges was allowed. The variable considered was speech articulation, which was defined as efficacy of sound production for intelligibility of oral communication and was considered *precise* (when all sounds were well defined and clear) or *imprecise* (when at least some sounds were not well defined and could not be fully understood).

Acoustic analysis

According to previous research[24], spontaneous speech (monologue) was used for acoustic analysis since it can be considered suitable for the evaluation of speech articulation in PD. Speech samples were digitally recorded and anonymized by a speech therapist (AED) in a quiet room using Shure microphone SM-58, placed approximately 15 cm from the patient's mouth. The formant frequency values F1 and F2 were measured separately for each vowel for a 30-millisecond segment at the temporal midpoint, determined by three blinded examiners, of each vowel using a PRAAT software v5.3.30 [available at: www.praat.org (Phonetic Sciences, University of Amsterdam)].

The vowel analysis was performed using the spontaneous speech in which the patients were instructed to speak about their speech difficulties. For each patient, 10 occurrences of the three vowels /a/, /i/ and /u/ were extracted from de monologue and was based upon the established Vowel Articulation Index (VAI) develop by Sapir and coworkers[25]. The measurement of VAI can be expressed using the following formula: $(F_2/i/+F_1/a//)/(F_1/i/+F_1/u/+F_2/u/+F_2/a/)$, which is a surrogate parameter of the first and second formant

frequencies (F1 e F2) of the corner articulation vowels /a/, /i/ and /u/[26,27]. The vowel data of F1 and F2 in Hertz (Hz) were separately averaged for all corner vowels of each patient.

Acoustic analysis was performed separately for both genders because mean VAI is related to the speaker's fundamental frequency of voice.

Statistical analysis

Analysis of group distribution showed that data from this study do not follow a normal distribution and thus a non-parametric analysis was employed. The following non-parametric tests were performed: the Mann-Whitney test was employed to compare stage of disease, scores of UPDRS-III and speech articulation (acoustic and perceptual) between the two groups. The correlation analysis of Spearman was performed for the correlation of the same variables within each group.

Analysis reliability was estimated by the concordance degree among judges by Kappa coefficient. Concordance was established by comparing evaluations obtained by each jugde by Cronbach α correlation ($p \leq 0.05$).

Ethics

The study was submitted to the Commission of Ethics for the Analysis of Research Projects (CAPPesq) and approved. All participants were previously fully informed about the research and signed consent forms before they were submitted to any evaluation.

RESULTS

Table 1 shows sample characteristics and the comparisons between groups regarding neurological and articulation features. Both groups were homogeneous in relation to disease duration, clinical stage, motor status and perceptual and acoustic behavior of speech articulation. Overall, these results indicate that PD patients with different ages at onset were clinically similar.

Group I included patients between 48 and 69 years of age and age of onset between 42 and 55 years group II included patients between 68 and 94 years of age and age of onset between 65 and 84 years.

Both groups had similar disease duration (2-18 years) and HY staging (between 2 and 4). Motor scores according to the UPDRS scale and speech articulation behavior were not statistically significant between groups.

Table 2 shows perceptual characteristics of speech articulation.

Table 3 shows correlations between neurological and perceptual and acoustic articulation characteristics in both groups. Statistical analysis showed a positive relationship between perceptual ratings and acoustical measures and severity of speech impairment and axial and bradykinesia scores.

Kappa correlation analysis indicated significant intra-judge concordance for perceptual and acoustic evaluation (0.879; $p \leq 0.01$). Cronbach α correlation analysis indicated significant inter-judges concordances for perceptual and acoustic evaluation (0.957; $p \leq 0.01$).

Table 1. Demographics, clinical presentation and articulation characteristics.

Characteristics	Group I (n = 30) Middle-age onset Mean ± SD	Group II (n = 20) Old-age onset Mean ± SD	Significance: p ≤ 0.05 Mann-Whitney test
Age	59.96 ± 4.85	80.40 ± 7.37	p < 0.001
Age at onset	49.26 ± 3.41	70.85 ± 7.33	p < 0.001
Duration of disease	10.56 ± 4.46	9.73 ± 4.43	p = 0.538
Hoehn and Yahr	2.71 ± 0.71	3.18 ± 0.75	p = 0.736
UPDRS			
Tremor	4.66 ± 2.68	4.15 ± 2.71	p = 0.611
Rigidity	9.80 ± 2.96	9.05 ± 3.15	p = 0.544
Bradykinesia	14.70 ± 4.41	15.45 ± 5.96	p = 0.506
Axial	11.43 ± 5.25	13.45 ± 5.32	p = 0.190
Perceptual analysis			
Articulation	0.43 ± 0.50	0.50 ± 0.51	p = 0.528
Acoustic analysis			
VAI – Male	0.75 ± 0.09	0.79 ± 0.05	p = 0.260
VAI – Female	0.81 ± 0.07	0.88 ± 0.08	p = 0.492

UPDRS: Unified Parkinson's Disease Rating Scale; VAI: Vowel Articulation Index; SD: standard deviation.

Table 2. Perceptual characteristics of speech articulation.

Characteristics	Group I (n = 30) Middle-age onset N (%)	Group II (n = 20) Old-age onset N (%)	Significance: p ≤ 0.05 Mann-Whitney test
Articulation			
Precise	17 (57)	6 (30)	p = 0.114
Imprecise	13 (43)	14 (70)	p = 0.528

Table 3. Correlation between neurological symptoms and speech articulation.

Characteristic	Correlation coefficient	Significance: $p \leq 0.05$ Spearman analysis
Speech articulation (perceptual ratings x acoustical measures)	+ 0.165	$p < 0.001$
Speech articulation x UPDRS-III Axial	+ 0.350	$p = 0.013$
Speech articulation x UPDRS-III Tremor	+ 0.041	$p = 0.779$
Speech articulation x UPDRS-III Rigidity	+ 0.263	$p = 0.065$
Speech articulation x UPDRS-III Bradykinesia	+ 0.277	$p = 0.005$
Speech articulation x Hoehn & Yahr	+ 0.246	$p = 0.006$

UPDRS: Unified Parkinson's Disease Rating Scale.

DISCUSSION

Although aging itself is not a cause of PD, the disease is an age-related disorder and affects all different aspects of speech and voice and leads to imprecise articulation of consonants and vowels[28].

In order to identify a possible relationship between age at onset and PD speech impairment, we analyzed the pattern of speech disturbances in individuals with middle-age and old-age onset PD. Both groups were homogeneous and there was no significant difference between groups for disease duration, HY stage, UPDRS-III scores and speech articulation (perceptual and acoustic). This suggests that the influence of age at onset on demographics data, clinical presentation and speech articulation was minimal or none.

Patients in our study were in the moderate or advanced stages of disease (mean disease duration of 10 years, average HY = 3). Our data indicate that the degree of articulation impairment is positively correlated with disease progression.

The specific pattern of the development of speech symptoms with disease progression is still unknown. Although we could not find any differences in overall UPDRS part III between groups, correlation analysis suggested that the severity of axial symptoms and bradykinesia were associated with poor and imprecise speech articulation performance in both groups.

In a study comparing acoustic speech characteristics and global motor performance a similar correlation with bradykinesia was found[18].

These similarities might be explained by shared pathophysiological mechanisms responsible for reduced articulation, bradykinesia and axial symptoms. Overall, these findings support the hypothesis that speech articulation impairment could be the result of axial dysfunction and bradykinesia.

We were able to document a speech articulation impairment not only based upon perceptual judgment, but also substantiated by objective acoustic measures with a VAI, an acoustic metric widely used to quantify articulatory function, as indication for a deterioration of articulation. The combination of perceptual and acoustic analysis of speech articulation as performed in the current study seemed to be appropriate to complimentarily obtain clinical surrogate measures of the articulation, and to measure changes that would be too subtle to be detected by perceptual judgment only. In our study, all patients presented preserved speech intelligibility, but alterations of vowel articulation were detected by a measurement of VAI with an average of less than 1.0 as indicator of articulation impairment[29].

Comparison of acoustic variables in relation to gender was not a primary concern but acoustic analysis was evaluated separately for both genders since mean VAI is related to fundamental frequency[26].

The presence of tremor may negatively influence some functional abilities such as writing and handling of common utensils but it does not appear to interfere with speech articulation. Rigidity could also influence speech since increased muscle tone may contribute to mobility limitation but in our study this correlation was not statistically significant. However, other studies suggest that bradykinesia and rigidity may contribute to the reduced mobility of speech-related structures and may play a role in the pathophysiology of impaired articulatory function[30].

Our study was not designed to characterize specific types of articulatory dysfunction or speech coarticulation. However it was possible to identify patterns of imprecise articulation and phonetic alterations, which included omission (when certain sounds were not produced), distortions (wrong emissions) and repetitions (when one or more sounds were duplicated in the same word).

Admittedly, this study has some methodological limitations mainly derived by our attempt to analyze only spontaneous speech in objective evaluation. In fact, according to a previous study, complex tasks such as monologue are more likely to elicit articulatory deficits in parkinsonian speech[24]. The impact of the age of onset on speech articulation cannot be captured by acoustic and perceptual analysis alone and disability or functional changes perceived by the patients themselves and/or their caregivers should also be taken into account.

In conclusion, our study suggests that global motor disability and speech articulation impairment do not correlate with age at onset of PD symptoms or age of the patients at evaluation. Moreover, speech impairment was associated with axial symptoms, bradykinesia and stage of the disease.

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