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Motor speech disorders and dysphagia in neurodegenerative diseases: clinical profiles compared

The aim of this study is to detect the alterations in the acoustic parameters of speech and swallowing difficulties in neurodegenerative diseases. 14 patients affected by Parkinson's Disease, 5 patients affected by Atypical Parkinsonism and 8 patients affected by Multiple Sclerosis were enrolled and underwent clinical and instrumental evaluation of speech and swallowing. The data obtained have been compared to those of a control group of 15 healthy people. The articulatory abilities have been assessed through maximum phonation time (MPT) and triangular voice space area (tVSA). Significant differences have been found between clinical and control group in MPT and tVSA. Regarding the swallowing assessment, parkinsonian syndromes showed worse results than Multiple Sclerosis in terms of bolus stagnation. In the patients group, speech and swallowing disorders are not similar in terms of neither severity nor onset. Noticeably, in our corpus dysphagia never represents the only disorder, as patients with dysphagia also show speech impairment.

Keywords: dysarthria, dysphagia, neurodegenerative disease, speech production.

1. *Introduction*

Dysarthria and dysphagia are common symptoms in neuromuscolar disease (Müller, Wenning, Verny, Mckee, Chaudhuri, Jellinger & Litvan., 2001; Nishio, Niimi, 2004; Perez-Lloret, Nègre-Pagès, Ojero-Senard, Damier, Destée, Tison, & Copark Study Group, 2012; Knuijt, Kalf, De Swart, Drost, Hendricks, Geurts., Van Engelen, 2013). 2013; Wang, Carter & Altman, 2020). Dysarthrias are speech disorders that result from neurologic impairments associated with weakness, slowness, or incoordination of the musculature used to produce speech. These speech disorders occur with considerable frequency in individuals with Parkinson disease, stroke, cerebellar disease, amyotrophic lateral sclerosis, multiple sclerosis, cerebral palsy, and traumatic brain injury (Duffy, 1995; McNeil, 1997; Kent, 2000). Neurogenic dysphagia is defined as an impairment of the swallowing ability that propels the food from oral cavity to the stomach and protects airway from food, liquids, and other substances in patients with neurological disease of various etiologies (Panebianco, Marchese-Ragona, Masiero & Restivo 2020). In literature, dysarthria appears to be a strong clinical indicator for the presence of dysphagia, and the severity of dysarthria seems to correspond to severity of dysphagia (Malandraki, Hind, & Gangnon, 2011; Ko, Chae & Cho, 2018; Wang *et al.*, 2020). The prevalence and severity of dysphagia varies depending on the type of dysarthria (Nishio *et al.*, 2004; Wang *et al.*, 2020).

The more severe conditions of dysphagia seem to be linked to dysarthria associated with bilateral impairment of upper or lower motor neurons such as flaccid, spastic, and mixed dysarthria. The idea that dysarthria and dysphagia could be correlated based on degree of severity is also supported in a 2010 study where the most severe dysarthria cases were associated with oropharyngeal dysphagia (Otapowicz, Sobaniec, Okurowska-Zawada, Artemowicz, Sendrowski, Kułak, Kuzia-Śmigielsk, 2010; Wang *et al.*, 2020). However, the correlation between dysarthria and oral-oropharyngeal dysphagia remains unclear (Wang, 2020). The aim of this study is to detect the presence of alterations in the acoustic parameters of the voice and swallowing function to outline clinical features of neurodegenerative diseases.

2. *Materials and methods*

The study included 14 people with Parkinson's disease (PD), 5 with atypical parkinsonism (PA) and 8 with multiple sclerosis (MS) enrolled at the at Ear, Nose and Throat Unit of University Hospital in Bari. Patients with other neurological or otolaryngological conditions that could affect swallowing or phono-articulation abilities were excluded from the study. Patients with Parkinson's disease were assessed using the Hoehn & Yahr clinical staging system (Hoen & Yahr, 1967), and patients with multiple sclerosis were assessed using the Expanded Disability Status Scale (Bushnik, 2011). The clinical group is 70 % composed by Parkinsonian patients and 30 % by Multiple Sclerosis patients. In the Parkinsonian group, the prevalence of the pathology in males is 58 % with a greater representation of the seventh decade of age. On average, the H&Y score is 2.8. Time since diagnosis (TSG) averages 7 years. The sample of people affected by Multiple Sclerosis is equally divided between the two sexes with a greater representation of the fourth decade of age. On average, the EDSS score is 4 (0.3). The TSG averages 20 years. Table 1 provides a summary of the demographic characteristics of the sample studied and the control group.

Table 1- *Demographic characteristics of clinical and control group*

	PD+PA	MS	CG
N (%)	19 (70)	8(30)	15
Mean age (±SD)	66.9 (±11.6)	50 (±11.2)	55 (±4.4)
Sex			
Male (%)	11 (58)	4 (50)	6 (40)
Female (%)	8 (42)	4 (50)	9 (60)
TSD (±SD)	7 (±3.6)	20 (±12.2)	
Stage of disease	2.8 (1.3) H&Y	4.1 (0.3) EDSS	

Each patient underwent clinical and instrumental swallowing evaluation and phonoarticulatory evaluation using acoustic analysis of voice. Control group subjects only underwent phonoarticulatory evaluation.

Statistical analysis of the data obtained was performed with the Software SPSS (IBM Corp, 2022): the T-student test, variance and covariance analysis were used.

2.1 Phono-articulatory evaluation

Voice recording was performed using a Samson Meteor Mic – USB Studio Condenser Microphone (frequency response of 20Hz-20kHz) placed 20 cm from the lips in a quiet room (30 dB background noise) and connected to the Audacity 2.1.2 software. The recording was done in mono channel with a sampling rate set at 44 kHz and 16 bits. The input volume was set to 70 %. The protocol proposed by Vizza, Tradigo and Mirarchi (2019) was used in the present study.

To evaluate the patients articulatory abilities, areas of the vowel triangles were analyzed. Each patient was instructed to perform a continuous and sustained vowel /a:/ to evaluate the Maximum Phonation Time. Patients able to pronounce a vowel for more than 5 seconds were asked to produce continuous and sustained vowels /a:/, /i:/, /u:/ for this duration. Subsequently, the triangular vowel space area of sustained vowels (tVSAv) was calculated using the F1 and F2 values of the sustained vowels /a:/, /i:/, /u:/. Subsequently subjects were asked to read a phonetically balanced text; the triangular vowel space area of semi-structured speech (tVSAs) was calculated using the average F1 and F2 values of three vowels /a:/, /i:/, /u:/ preceded and followed by plosive consonants in the text.

The vocal signal was analysed using Praat (Boersma, P. & Weenink, D., 2021). Areas were calculated using GeoGebra (Hohenwarter *et al.*, 2020).

2.2 Swallowing evaluation

Swallowing evaluation was carried out by clinical and instrumental assessment and was preceded by the administration of a self-report questionnaire. The Dysphagia Risk Assessment Scale (DRAS) questionnaire was used (Fukada, Kamakura & Kitaike, 2013; Cavallaro, Pontrelli & Sammali, 2022). The clinical assessment was carried out using the Bedside Swallowing Assessment Scale (BSAS), a scale that is useful in assessing the presence of the conditions necessary to start swallowing, the presence of defence mechanisms and the patient's ability to swallow using water testing. The instrumental assessment was then carried out by means of a fibroendoscopic examination of swallowing (FEES). The morphology and motility of the pharyngo-laryngeal structures and the search for possible salivary stagnation were assessed prior to the swallowing tests. The presence pre swallow spillage, penetration and aspiration, voluntary and reflex coughing, and post swallow stagnation in the valleculae and pyriform sinuses were observed at liquid (60 mL of water), semi-solid (yoghurt) and solid (dry biscuit) bolus test. Finally, the laryngeal adductory reflex (LAR) was assessed stimulating the arytenoid surface. The stagnation severity was assessed using the Yale Pharyngeal Residue Severity Rating Scale (Neubauer, Rademaker & Leder, 2015), which distinguishes between severe, moderate, mild, trace or absent stagnation. A score from 1 to 7 was obtained

on the Dysphagia Outcome and Severity Scale (DOSS) based on the impairment detected at the FEES.

3. *Results*

3.1 Swallowing evaluation results

Most patients included in the Parkinsonian group showed a mild to severe dysphagia with a DOSS score less than or equal to 5: 55 % of patients had a score equal to 5, 7 % of patients had a score equal to 4, 10.5 % of patients had a score equal to 3, 10.5 % of patients had a score equal to 2. The parkinsonian group showed an average DOSS score of 4.6, corresponding to a tendentially mild to moderate degree of dysphagia.In the Parkinsonian group, the average score obtained on the DRAS questionnaire is 14.

Clinical and instrumental evaluation of swallowing function showed pathological GAG-reflex in 53 % of cases, pathological voluntary cough in 32 % of cases, pathological LAR in 37 % of cases. Saliva stagnation was shown by 32 % of patients in the valleculae and by 21 % of patients in the pyriform sinuses. At liquid bolus tests, 42 % showed stagnation in valleculae and 44 % showed stagnation in pyriform sinuses. At semi-solid bolus tests, 56 % of patients showed stagnation in the valleculae and 50 % showed stagnation in the pyriform sinuses. At solid bolus tests 53 % of patients showed vallecular stagnation, 35 % showed stagnation in the piriform sinuses. No penetration was observed; aspiration was observed in 2 patients (11 %).

Patients included in the Multiple Sclerosis group showed a mild to moderate dysphagia: 29 % of patients had a score equal to 5, 5 % of patients had a score equal to 4. The average DOSS score was equal to 5.6, corresponding to mild degree of dysphagia. In the Multiple Sclerosis group, an average score of 12.2 was obtained from the DRAS self-assessment questionnaire.

Clinical and instrumental evaluation of swallowing function showed pathological GAG-reflex in 38 % of cases, pathological voluntary cough in 13 % of cases, pathological LAR in 38 % of cases. Saliva stagnation in the valleculae was observed in 13 % cases. At semi-solid bolus tests, 25 % of patients showed stagnation in the valleculae and 13 % showed stagnation in the pyriform sinuses. No stagnation was observed during liquid and bolus trials.

3.2 Speech evaluation results

The 100 % of patients included in the Parkinsonian group had a TOM score less than or equal to 4 showing a mild to severe dysarthria: 35 % of patients had a score equal to 4, 28 % of patients had a score equal to 3, 21 % of patients had a score equal to 2, 10 % of patients had a score equal to 1, 7 % of patients had a score equal to 0. The parkinsonian group showed an average TOM score of 2.7, corresponding to a tendentially moderate degree of dysarthria. The Maximum Phonation Time (MPT) has an average duration of 11s; 36 % of patients have a pathological phonatory

duration, ranging from 0s to 10s. The prevalent MPT is in the range of 9.1s -19s. The average value tVSAv is 145,739 Hz²; the average tVSAs is 112,648 Hz².

In the Multiple Sclerosis group, 57 % of patients showed a mild to severe dysarthria: 75 % of them had a score equal to 4 and the remaining 25 % of patients had a score equal to 3. The average TOM scale score of 4 corresponding to a mild degree of dysarthria. MPT has an average duration of 10.83 s. The average value of tVSAv is 265,835 Hz²; the average tVSAs is 157,094 Hz².

In the control group, MPT has an average duration of 23 s. The tVSAv average value is 260766 Hz² and the tVSAs average value is 289268 Hz².

Statistically significant difference was found between tVSAv and tVSAs in the clinical group (p=0.04) and borderline statistical significance between tVSAs in MS and control group (p=0.05). A borderline significant difference was found between the TMF of the MS patients and the control group (p=0.05). The analysis of covariance was used to study the trend of tVSAs, taking into account tVSAv as a covariate. A significant difference was found between the tVSAs values of controls compared to those of people with PD (p=0.0002), atypical PD (p=0.0082) and MS (p=0.001).

In the Parkinsonian group dysarthria and dysphagia coexisted in 83 % of cases, the remaining 17 % of patients only showed dysarthria. In the Multiple Sclerosis group 29 % of patients did not show either dysarthria or dysphagia, 38 % of patients only showed dysarthria and 14 % of patients only showed dysphagia. Swallowing and phono-articulatory disorders coexisted in 19 % of patients.

4. *Discussion*

The assessment of swallowing and phono-articulatory function made it possible to outline the clinical features of the pathologies under examination. Changes in the pneumophonic system, such as reduced respiratory support or glottic insufficiency, may explain the differences between the MPT in the clinical and in the control group.

Using tVSA for assessing speech intelligibility turned out to offer interesting results. The differences found between the control and clinical groups could be led to changes in the amplitude, strength and coordination of the movements required to produce phonemes. Within the clinical groups, it is observed a discrepancy between the values of the tVSA obtained from semi-structured speech and those obtained by extracting the formant values from sustained vowels. Indeed, the areas calculated on sustained vowels are significantly larger than those extracted from spontaneous speech. These results support the use of the vowel polygon extracted by more ecological tests to assess speech intelligibility. In the clinical group, most of the times both dysphagia and dysarthria were prevalent but speech and swallowing disorders were not similar either in terms of severity or onset. Atypical parkinsonism, which had the worst swallowing and phono-articulation scores, had the most severe clinical phenotype.

There was also a difference between the subjective ratings of the patients in the study and the ratings of the patients in the control group. Specifically, patients with Parkinson's disease and atypical parkinsonism showed a lower susceptibility to dysphagia, even when the dysphagia was overt. Multiple sclerosis patients, on the other hand, reported more frequently swallowing difficulties, even when undetectable by instrumental evaluation. As self-report is not always a reliable measure, it is recommended to assess swallowing abilities even in the absence of reported symptoms, especially in PD.

This finding corroborates the hypothesis that dysphagia should be always investigated in neurodegenerative patients affected by motor speech disorders.

5. *Conclusions*

It has been illustrated the swallowing and phonoarticulatory performance profiles in patients affected by Parkinson's Disease, Atypical Parkinsonism and Multiple Sclerosis, comparing the data obtained with those coming from a control group.

In the clinical group, most of the times both dysphagia and dysarthria were prevalent. The next goal is to continue the study by expanding the sample size, using more consistent data, which would allow to apply more valid statistics and produce results which are more representative of patients with neurodegenerative diseases.

Acknowledgments

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