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Acquired non-specific stuttering in Parkinson's disease: a case report

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Sažetak

Parkinsonova bolest (PB) - progresivna je neurodegenerativna bolest koju karakteriziraju tremor, bradikinezija i rigor. Uz motoričke i nemotoričke manifestacije Parkinsonove bolesti, niz je drugih simptoma, uključujući poremećaje govora i druga kognitivna oštećenja. Najčešći simptomi govora su bradilalija, dizartrija, hipofonija i promijenjena prozodijska obilježja. Kognitivne promjene, koje se javljaju u prodromalnoj fazi PB-a, uključuju oštećenje izvršnih funkcija i radne memorije, nakon toga slijede teškoće pažnje i verbalne fluentnosti prije nego što motoričke teškoće PB postanu vidljive. Cilj istraživanja je prikazati slučaj 74-godišnje bolesnice s Parkinsonovom bolešću, koja ima govorno-jezične poteškoće i atipičnu govornu netečnost. Dijagnostička obrada provedena je pomoću kliničke baterije testova za jezično-govornu i neuropsihološku procjenu. Rezultati jezično-govorne procjene ukazuju na značajno smanjenu govornu razumljivost zbog netečnosti, distorziranu artikulaciju, teškoće u organizaciji spontane ekspresije i razumijevanja gramatičkih struktura, narušenu verbalnu fluentnost i teškoće u receptivnom vokabularu. Neuropsihološka obrada ukazala je na difuznu deterioraciju ispitivanog kognitivnog funkcioniranja, veću od očekivane, uzimajući u obzir kronološku dob i vjerojatno dobre premorbidne sposobnosti.
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Ključne riječi:
Parkinsonova bolest; tečnost govora; mucanje; jezične poteškoće; terapija

Summary

Parkinson's disease (PD) is a progressive neurodegenerative disease predominantly characterized by tremor, bradykinesia, and rigor. In addition to motor and non-motor manifestations of Parkinson's disease, there are a number of symptoms, including speech disorders and other cognitive impairments. The most common speech symptoms are bradylalia, dysarthria, hypophonia and impaired prosody. Cognitive changes that occur in the prodromal phase of PD include impairment in executive functions and working memory, followed by impairment in attention and verbal fluency, and that is before the motor characteristics of PD become visible. The aim of the study is to present the case of a 74-year-old patient with Parkinson's disease who has speech and language difficulties and atypical speech disfluency. Diagnostic processing was performed using a clinical battery of tests for speech – language assessment and neuropsychological assessment. The results of the speech – language assessment indicate significantly reduced intelligence due to non-specific speech disfluency and inaccurate articulation, difficulty in organizing spontaneous expression and understanding grammatical structures, impaired phonemic verbal fluency and difficulties in receptive vocabulary. Neuropsychological processing indicated diffuse deterioration of the examined cognitive functioning to be larger than expected when taking ito consideration the age and probably good premorbid abilities of this person.

Key words:
Parkinson's disease; speech fluency; stuttering; language difficulties; therapy.

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INTRODUCTION

Parkinson's disease (PD) is a progressive neurodegenerative disease caused by the degeneration of dopaminergic neurons, leading to clinical features predominantly characterized by tremor, bradykinesia, and rigor (Im et al., 2018, p. 150). On the other hand, there are atypical parkinsonian syndromes (APS), such as progressive supranuclear palsy (PSP) and multiple system atrophy (MSA), which differ from PD by more widespread neuronal involvement, resulting in additional clinical signs, more rapid disease progression and poor response to dopamine replacement therapy (Rusz et al., 2015, p. 993). In addition to motor and non-motor manifestations of Parkinson's disease, there are a number of symptoms, which include speech disorders and other cognitive impairments. The most common speech symptoms are bradylalia, dysarthria, hypophonia and impaired prosody, and in the advanced stage of the disease, dysphagia is often encountered (Brown and Spencer, 2020, p. 2145). Cognitive changes that occur in the prodromal phase of PD include impairment in executive functions and working memory, followed by impairment in attention and verbal fluency, and that is before the motor characteristics of PD become visible. Executive functions refer to a multidimensional process that includes thinking (flexibility, abstractness), planning and goal-oriented behavior, self-control and inhibition, will and sustainability of attention, and their disruption leads to psychological and social dysfunction. Deficiency of executive functions occurs both in the early and late phase of the disease, while in the late phase there is also deficit of visuospatial abilities. Early and accurate diagnosis is essential for understanding the underlying pathophysiology, in assessing prognosis and making decisions regarding treatment (Juste, Sassi, Costa, de Andrade, 2018; Brown and Spencer, 2020, p. 2146). Speech disorder is a common clinical manifestation occurring in 70–100% of patients with PD, PSP and MSA (Ho, Iansek, Marigliani, Bradshaw, Gates, 1998, p. 132) and tends to develop at an early stage (Rusz, Cmejla, Ruzickova, Ruzicka, 2011, p. 351). Whilst the majority of PD patients develop a clear form of hypokinetic dysarthria (Ho et al., 1998, p. 133), PSP and MSA patients typically develop mixed dysarthria with various combinations of hypokinetic, spastic and ataxic components (Kluin, Gilman, Lohman, Junck, 1996, p. 547) due to the involvement of the basal ganglia, corticobulbar pathways and the cerebellum. Analyzes of motor speech disorders can provide important indications for the diagnosis and pathophysiology of the underlying disease (Rusz et al., 2015, p. 994), especially since speech assessment is a cheap, non-invasive and rapid technique compared to neuroradiological techniques that could potentially be used in evaluating subjects with initial Parkinsonism (Rusz et al., 2011, p. 359).

The aim of the study is to present the case of a 74-year-old patient with Parkinson's disease with speech and language difficulties and atypical speech disfluency.

CASE REPORT

A 74-year-old female patient with Parkinson's disease came to the Center of SLP and Rehabilitation Blaži with a speech difficulty and inability to write. The patient gave written consent to present her case. These difficulties have intensified in the last six months, which is why the patient has a subjective feeling that she can no longer speak normally, and phone conversations are especially difficult for her. Consequently, she communicates verbally only with close family members (with sister, brother and nephew). She states that she knows exactly what she wants to say, but it is difficult for her to speak. On the other hand, the patient and her accompanying nephew state that when she speaks German, the mentioned speech difficulties are not so frequent. She notices that her voice is hoarse, and she loses her breath while eating. She denies frequent coughing. She generally feels tired. She mentions problems with facial expressions, i.e., changes in facial expressions during exercises which she performed with a speech therapist in Germany. When speaking, she sometimes finds it harder to find the right word. People often ask her to repeat what she said, that is, they find it difficult to understand her. In moments of pronounced speech difficulties, she cannot stop and perseveres, often letting others speak in her name. She feels excluded from the conversation because she can no longer speak as before. Her difficulties result in a slight sense of shame. She was diagnosed with Parkinson's disease in 2011 in Germany, where she had lived until 2017. In Germany, she was involved in certain rehabilitation procedures, and one of them was speech therapy. She also attended speech therapy in Karlovac for a short time, but medical reports from Karlovac were not available to us. Until the beginning of the pandemic caused by Covid-19 disease, she was controlled by a neurologist, cardiologist, nephrologist and nuclear medicine specialist in Germany. Given the impossibility of further visits to Germany due to the epidemiological situation, the first examination by a neurologist in Croatia was performed in January 2021 at the Polyclinic Croatia. In addition to Parkinson's disease, this patient also has verified arterial hypertension, hypothyroidism and hyperlipidaemia, mitral regurgitation and chronic ischemic heart disease due to non-rheumatic mitral valve dysfunction and ventricular extrasystole. The patient also underwent a total thyroidectomy for the treatment of verified thyroid cancer (she did not provide medical documentation). Her drug therapy includes Euthyrox, Metoprolol, Atorvastatin, Rasagiline, Pramipexol and Madopar (according to the medical report from January 2021, the doses are as follows: Razagilin a 1 mg 1.0.0, Pramipexol a 0.7 mg 3x1, Madopar pill 3x1) On the last neurological examination and processing, registered speech disorders, hypomimia and paresis of vertical vision were evaluated as mediocre, while the function of other cranial nerves was adequate, and the diagnosis of Parkinson's disease was confirmed.

Neuropsychological assessment was performed at the Department of Neurosurgery, University Hospital Center Zagreb, and the results indicate cognitive functioning at the level of MCI. The assessment included a

clinical interview and the following diagnostic inventory: Raven's Coloured Progressive Matrice (CPM) (Raven, 1998), Auditory Verbal Learning Test (Rey, 1964), The Rey-Osterrieth Complex Figure Test and Recognition Trial (Osterrieth, 1944), Wechsler Adult Intelligence Scale (WAIS) (Wechsler, 1986) (wbII-sb; Subtests: Information – General Information Preservation of Old Factual Knowledge, Comprehension, Arithmetic, Digit Span – forward and backward, Similarities, and Vocabulary, Picture Arrangement, Picture Completion, Block Design, Object Assembly, and Digit Symbol), Trail making test (TMT) for executive functions as well as clock drawing test (CDT), MMSE subtests (The Mini-Mental State Exam; Subtests - temporal orientation, spatial orientation, immediate memory, attention/ concentration, delayed recall, verbal repetition (1 point), verbal comprehension, writing, reading a sentence), related to time orientation and graphomotor perceptual abilities and writing. Neuropsychological processing indicated diffuse deterioration of the examined cognitive functioning larger than expected when taking into consideration the age and probably good premorbid abilities of this person. Severe disorders of speech and language functioning, constructive dyspraxia and disorders of visuospatial abilities dominated. Mnestic functions were still good for age, short-term verbal memory was very good, while long-term verbal and working memory had borderline deviations that are suspected be of the combined type.

Recent speech therapy assessment and diagnostics were conducted with the aim of evaluating the overall language and speech status and reading and writing skills. The following diagnostic instruments were used in the assessment: clinical interview, Test for Reception of Grammar (TROG-2: HR) (Kuvač Kraljević, Hržica, Kovačević, Kologranić Belić, 2013), Peabody Picture Vocabulary Test (PPVT-III-HR) (Kovačević et al., 2009), Articulation Test (Vuletić, 1990), Oral-Motor Ability Test tasks, Battery of Language Competences Isolated Test tasks (naming tasks, automated series, orientation, phonological naming and processing, linguistic comprehension of orders and abstract concepts), tasks for testing reading and writing skills (non-standardized, clinically proven form). Additionally, questionnaires were used for self-assessment of swallowing, voice/speech, and salivation assessment (internally translated ROMP questionnaires). The patient came to the speech therapy examination on her own, using a medical walker. During the examination, she was motivated, cooperative, had adequate contact distances and was focused on the test material, although fatigue was noticeable after some time, as well as weakening of the body posture, which is why it was necessary to remind her to look up. Upper extremity tremor, bradykinesia, and hypomimia were noted. The voice was assessed by subjective evaluation. The voice was asthenic, reduced in volume, and hoarse. Occasionally, there were motor accompanying movements (sucking the thumb / biting the nails, putting finger in the nose, biting the cheeks, movements of the lower jaw, closing the eyes), as well as perseverations of movement. Based on a questionnaire for self-assessment of swallowing, voice and speech, and assessment of salivation, the patient denied difficulty swallowing, but stated extended feeding time and meal duration. She denied hypersalivation but feels the accumulation of saliva in her mouth. She noticed that her voice sounds rougher than before. She stated that she is often incomprehensible to familiar people, especially when she is tired, while she is even more incomprehensible to strangers and has to repeat what she wants to say over and over again, as well as when she uses the phone. She also stated that when she starts speaking, she needs help to put her thoughts into words. She can participate in a group conversation if she is helped by familiar people. She mentioned speech difficulties as the most difficult aspect of the disease. She achieved an extremely low result on the Test for Reception of Grammar (SR 55; 1st percentile). Qualitative analysis of the results indicates the presence of lexical and occasional errors, which further indicate difficulties in language processing. Also, she occasionally repeated additional tasks, which can indicate memory difficulties and slower language processing, and fatigue was noticed in the final repetitions. She scored moderately low on the Peabody Picture Vocabulary Test (PPVT-III-HR) (SV 75). The assessment of the oral-motor abilities of the articulator showed reduced static and dynamic power of the articulator, especially the tongue, reduced amplitude of the articulator's movement and poorer coordination. Ingestion was prolonged in all three phases. In case of continuous swallowing, due to fatigue, she was taking breaks. After a short period, aspiration occurred intermittently with a consequent cough and a change in the quality of the timbre of the voice (wet, gargling voice occurred). The patient's phonation was asthenic, hoarse and noisy, with reduced volume and dynamic range, especially during prolonged talking, where laryngeal fatigue occurred. Phonation time was reduced. The speech was chanted and of a variable tempo. The patient reported that sometimes she cannot control her speech, and sometimes she deliberately hurries to make it easier to speak. During the assessment, pronounced specific and non-specific speech disorders, which significantly impair the intelligibility of speech and make verbal communication with the environment more difficult, were noticed. An intertwined network of spoken and non-spoken behavior was visible in the form of speech perseverations with difficult speech in the form of multiple fast and inconsistent repetition of parts of words/voices and prolongation. Sometimes it took a long time to inhibit these difficulties. She often returned to the beginning of the word at the moment of inaccuracy, for the purpose of trying to continue the speech, but mostly without success. Also, the difficult pronunciation of individual voices was visible, especially palatal affricates. The patient is aware of these difficulties and verbalizes them herself. Accompanying movements (benign eyelid microclones, facial muscle spasms and mandibular spasms) were registered during spontaneous expression in severe disorders, supported by manifested anxiety symptoms due to communication and social judgment situations. Giving up or avoiding speech during the assessment was not noticed. The mentioned speech symptomatology was present at all language-speaking levels, the least expressed in reading, repetition by model and in the production of automated series, while in spontaneous speech it was very pronounced (sometimes on almost every word in a sentence). Articulation was imprecise and distorted. She

performed automated series in both directions with a faster speech rate and occasional inaccuracies. Singing was also disrupted. Naming was performed with speech inaccuracies, and it seemed that less frequent and well-known terms and the need for explanation encourage speech difficulties. Difficulties were also present in verbal fluency. Repetition according to the model - words and sentences were mostly well executed, although when repeating longer words/sentences, the above-described speech difficulties occurred. She was well oriented spatially, temporally and towards people. The body scheme was clear. Spontaneous speech was organized and meaningful, but chanted and systematically very fluid, with inconsistent symptomatology of reduced intelligence and impaired prosodic features. Linguistic understanding of single and double orders was preserved, but unsystematically uncertain in complex utterances and abstract concepts. Automated reading was best preserved, except when reading multi-syllable words, when the described symptomatology occured. Comprehension of a short read text was good. Writing something down was not possible, as well as independent writing.

The patient has been included in speech therapy since September 2021 with the aim of improving oropharyngeal and speech-language functions. Speech therapy was primarily aimed at reducing severe speech difficulties, but also at targeted strengthening of all structures involved in speech production (speech breathing, articulator mobility, swallowing, phonation). Swallowing exercises and articulator mobility exercises were performed to strengthen muscles and improve control and precision of movements required for speech using the TalkTools program to stimulate/strengthen oral-motor skills. In order to improve speech and increase speech fluency, speech breathing exercises with an emphasis on taking a full breath before the beginning of speech were used, as well as techniques of using slower speech with prolonged beginning of speech. In the modification of speech, the levels moved from linguistically lower and very structured (automated series, repetition according to the model) to linguistically higher, lexically and syntactically more demanding statements. By using all of the above mentioned techniques, the patient unsystematically managed to be more fluent, i.e. speech difficulties were less present, but more frequent support was needed. In addition to verbal, visual support was used to facilitate the use of the above techniques. The material in the paper was often placed at eye level with the aim of strengthening proper posture of the body. Since reading skills are much more preserved, compared to speech expression, the above was also used to achieve more fluent speech while using the above techniques. In order to facilitate communication with the environment, the patient was introduced with and encouraged to use assisted means of communication (communication board), as well as additional strategies (more frequent use of nonverbal communication, use of shorter phrases, determining the topic before the interview, etc.).

DISCUSSION AND CONCLUSION

The analysis of previous studies shows that the characteristics of speech disorders may reflect the underlying neuropathology of PD and APS. Dysarthria was equally present in all patients with PSP and MSA and generally consisted of a combination of hypokinetic, spastic, and ataxic components, whereas patients with PD exhibited pure hypokinetic elements. Hypokinetic-spastic dysarthria predominates in the speech of patients with PSP, while predominantly ataxic dysarthria was manifested in patients with MSA, which probably reflects the high sensitivity of speech to minor cerebellar deficits. Disfluency is, according to literature, the only single speech aspect characteristic of PSP, but it is rarely observed in MSA and is usually associated with poor working memory, in contrast to stuttering-like behavior commonly observed in PSP and later stages of PD (Kim, Kent, Kent, Duffy, 2010, p. 68; Duffy, 2013, p. 378; Rusz, Cmejla, Tykalova, Ruzickova, Klempir, 2013, p. 2175; Rusz, Megrelishvili, Bonnet, Okujava, Brozova, 2014, p. 660; Saxena, Behari, Kumaran, Goyal, Narang, 2014, p. 858; Novotny, Rusz, Cmejla, Ruzicka, 2014, p. 1371). The onset of stuttering-like behavior may be due to the involvement of the globus pallidus and the primary motor cortex, which are regions of the brain commonly affected by PSP (Nath, Ben-Shlomo, Thomson, Lees, Burn, 2003, p. 913). Stuttering has usually been registered as a consequence of pallidal deep brain stimulation in patients with dystonia (Nebel, Reese, Deuschl, Mehdorn, Volkmann, 2009, p. 168), and is widely present in manganese (Ephedrone) - induced Parkinsonism associated with toxic and neurodegenerative damage to the globus pallidus (Rusz et al., 2014, p. 1372). It should be noted that motor planning is responsible for the control of speech fluidity coded in the left primary motor cortex, while this motor asymmetry of speech is missing in stuttering (Neef, Hoang, Neef, Paulus, Sommer, 2015, p. 713), but also, increased dopamine levels in PD can lead to stuttering with the motor cortex playing a similar role as in the case of levodopa-induced dyskinesia (Ostock, Dupre, Jaunarajs, Walters, George, 2011, p. 758; Tykalova, Rusz, Cmejla, Klempir, Ruzickova, 2015, p. 1140). Due to uncontrolled movements of the laryngeal muscles, fluctuations in vocal cord tension, and incomplete vocal cord closure, patients with MSA have poorer voice control compared to PSP, and changes in voice timbre quality, excessive pitch fluctuations, and vocal tremor may together give a perceptual impression of tense speech with trembling and elevated tone, while severe voice roughness in PSP subjects may resemble dysarthria. These aspects that contribute to reduced voice quality are rather nonspecific symptoms of neuronal dysfunction. The speech of patients with PSP is characterized by a slower rate followed by inappropriate pauses, more affected vowel articulation, longer phonemes and excessive variations in intensity, which may be due to striatum damage and generally more widespread neuronal atrophy (Hartelius, Gustavsson, Astrand, Holmberg, 2006, p. 245). The predominant hypokinetic-spastic dysarthria with fewer ataxic components may be due to extensive neurodegeneration involving the midbrain as well as the

globus pallidus, striatum, hypothalamic nucleus, pons, cerebellum and cerenucleum (Rusz et al., 2015, p. 998; Nath et al., 2003, p. 914) while the clinical features of dysarthria in MSA patients showing predominant ataxic dysarthria with fewer spastic and hypokinetic components are consistent with known neuropathological changes including cerebellar degeneration, midbrain, striatum, substantia nigra, and mandibular nucleus (Gilman et al., 2001, p. 674). The study by Rusz et al. (2015) supports the role of corticobulbar pathways and cerebellum in the development of mixed dysarthria in APS because the relationships between the severity of spastic components and bulbar or pseudobulbar manifestations, as well as between the severity of ataxic components and cerebellar signs were observed. When interpreting the results, it is necessary to take into account the long stay of the patient in another language-speaking area – PPVT. All of the above points to specific and non-specific speech disorders that are intertwined with the clinical picture of PD, but certain non-specific speech symptoms can be an important differential-diagnostic sign for neurologists.

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