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**Therapy of swallowing and speech problem in patients with progressive
supranuclear palsy**

Терапија проблема гутања и говора код болесника са прогресивном
супренуклеарном парализом

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Therapy of swallowing and speech problem in patients with progressive supranuclear palsy

Терапија проблема гутања и говора код болесника са прогресивном супренуклеарном парализом

SUMMARY

Introduction Progressive supranuclear palsy (PSP) is a rare form of neurodegenerative extrapyramidal disease. In addition to symmetrical parkinsonism, early falls, non-reactivity to dopaminemimetic therapy, the disease also manifests itself in swallowing problems with frequent choking and incomprehensible, difficult speech. In this paper, we present a patient with PSP who, in the clinical presentation of the disease, has a severe swallowing and speech disorder, in whom appropriate therapy was applied with a positive response of partial relief of the mentioned symptoms.

Case outline A 68-year-old male patient was referred to a speech therapist by a neurologist due to difficulties with swallowing and speaking. In the patient, the preparatory and oral phase of swallowing was impaired, with insufficient labial occlusion and weakened tongue mobility. The patient was involved in intensive speech therapy treatment for six months. The applied rehabilitation program resulted in the improvement of swallowing function and partial improvement of speech.

Conclusion The treatment of patients with PSP should be approached seriously and interdisciplinary, given the fact that there is no causal therapy available, and that the only option is symptomatic treatment of specific disabling conditions. It is essential to pay attention to the selection of rehabilitation programs that can improve speech and swallowing functions, as well as the quality of life for patients.

Keywords: progressive supranuclear palsy; swallowing; speech; rehabilitation

САЖЕТАК

Увод Прогресивна супрануклеарна парализа (ПСП) је редак облик неуродегенеративне екстрапирамидне болести. Поред симетричног паркинсонизма, раних падова, нереактивности на допаминиметску терапију, болест се манифестује и проблемима гутања са честим загрцњавањем и неразумљивим, отежаним говором. У овом раду приказујемо болесника са ПСП који у клиничкој презентацији болести има тежак поремећај гутања и говора, код кога је примењена одговарајућа терапија са позитивним одговором делимичног олакшања наведених симптома.

Приказ болесника Болесник мушког пола старости 68 година упућен је логопеду од стране неуролога због тешкоћа са гутањем и говором. Код пацијента је била нарушена припремна и орална фаза гутања, са недовољном лабијалном оклузијом и ослабљеном покретљивости језика. Болесник је био укључен у интензиван логопедски третман у трајању од шест месеци. Примењени рехабилитациони програм резултирао је побољшању функције гутања и делимичном побољшању говора.

Закључак Лечењу болесника са ПСП треба приступити озбиљно и интердисциплинарно, с обзиром на то да не постоји каузална терапија, те да је једина опција симптоматско лијечење специфичних онеспособљавајућих стања. Неопходно је обратити пажњу на одабир рехабилитацијских програма који могу побољшати функције говора и гутања, као и квалитету живота болесника.

Кључне речи: прогресивна супрануклеарна парализа; гутање; говор; рехабилитација

INTRODUCTION

Progressive supranuclear palsy (PSP) is the second most common form of neurodegenerative parkinsonism, after idiopathic Parkinson's disease [1], characterized by axial, levodopa unresponsive parkinsonism, with the presence of vertical gaze palsy, early postural instability with backward falls, cognitive and behavioral changes [2]. Otherwise, it is 10 times less common than typical Parkinson's disease. Some studies point out that an annual prevalence is 5 to 7 cases per 100,000 population [3], while the annual incidence rate ranges between 0.16 and 2.6 per 100,000 population [4].

According to the dominant clinical features, more than 10 subtypes of progressive supranuclear palsy have been described in the literature. The most common type is Richardson's syndrome (PSP-RS) which is characterized by early onset of postural instability, vertical supranuclear gaze palsy, and cognitive dysfunction. Otherwise, it is associated with a faster progression of the disease and a shorter survival time [5]. In contrast, the occurrence of other subtypes of PSP is very rare [6].

The average age of onset for disease is between 60 and 65 years, with a slight male predominance [7]. Some studies indicate that it is evenly distributed between males and females [8]. The average life expectancy in this form of PSP is 5 to 8 years [2].

The diagnosis of Progressive Supranuclear Palsy presents a significant challenge, especially in the early stages of the disease. In recent years, substantial progress has been made in clinical practice regarding the detection of this condition.

PSP is mainly levodopa unresponsive and up to now, no treatment strategy appears as successful. Therefore, the main treatment approach is symptomatic therapy.

We present a patient diagnosed with PSP, who exhibited pronounced swallowing and speech symptoms and underwent speech therapy rehabilitation. The implemented rehabilitation program led to improvements in swallowing function and a partial enhancement in speech.

CASE REPORT

The patient is a 68-year-old (retired) male, with no significant health issues, aside from high blood pressure. He has no known allergies and no positive family history of serious neurological disorders. The first symptoms appeared at age 62, when the patient began to notice difficulties with walking and speaking. Over time, these symptoms progressed and at the time of examination, at age 68, he presented with severe postural instability characterized by frequent backward falls, as well as slurred speech and difficulty swallowing.

A neurological examination revealed vertical gaze palsy and hypomimia. The patient exhibited dysarthric speech (slow, slurred, and poorly articulated) along with dysphagia. Bilateral bradykinesia and rigidity in the arms and legs were also noted. The applause test was positive. The patient's gait was slow and unstable, requiring assistance. There were no issues with sphincter control.

Basic laboratory tests were performed (including CRP, folic acid, thyroid hormones, vitamin B12, vitamin D and immunological tests). All laboratory parameters were within normal limits, except for low vitamin D levels.

Magnetic resonance imaging of the brain showed a "hummingbird" sign and a discreetly elevated signal of the tegmentum and tectum (Figure 1), as well as significant atrophy from the concave edge of the mesencephalon (Figure 2).

EEG findings are tidy. The patient underwent Holter monitoring and an EKG, both of which showed no deviations from normal. Cardiologically, only hypertension was diagnosed, and appropriate therapy was initiated. There were no signs of orthostatic hypotension.

The patient was diagnosed with probable progressive supranuclear palsy due to the presence of mandatory criteria and key characteristics including ocular motor dysfunction and postural instability. Additionally, the magnetic resonance findings supported the diagnosis of PSP [7]. The patient continued treatment with the correction of antiparkinsonian drugs.

Due to the impossibility of controlling food and liquids in the oral cavity, difficulty transferring bites or sips from the oral cavity to the pharynx, frequent choking and coughing, and then difficult and incomprehensible speech, the patient was referred for rehabilitation of speech and swallowing.

Assessment of swallowing and speech abilities

To evaluate swallowing and speech, we utilized the Water Swallowing Test [9], the Oral Practice Test [10], and the Global Articulation Test [11].

The water swallowing test is one of the screening tests we apply. It is a minimally invasive diagnostic procedure designed to identify oropharyngeal dysphagia. The patient is asked to sit down and drink 30 ml of room-temperature water from a glass in his hand as he normally does. During drinking process, the time taken to empty the glass is measured, and episodes of swallowing and drinking profile are monitored, which can indicate the need for further investigations. The drinking profiles that can be recorded are: Profile 1 – the patient can swallow all the water in one gulp without choking; Profile 2 – the patient can drink all the water in 2 or more sips without choking; Profile 3 – the patient can drink all the water in one gulp with occasional choking; Profile 4 – the patient can drink all the water in 2 or more sips with occasional choking; and Profile 5 – the patient often chokes and finds it difficult to drink all the water.

The Oral Practice Test assesses the voluntary muscle activity of the speech apparatus including the lips, cheeks, tongue, soft and hard palate and jaw. It evaluates the ability to perform specific voluntary movements of the orofacial muscles, which are controlled by certain cranial nerves (V, VII, IX, and XII). A disorder in any of these cranial nerves can lead to dysfunction in the orofacial region, resulting in impaired articulation. The test consists of 21 tasks, ranging from simple actions (such a breathing in and out through the nose, extinguishing a match), to very complex tongue movements (eg creating a palatolingual groove). Testing involves asking the subject to perform an oral-motor pattern demonstrated by the examiner. For each successfully completed task, the examinee receives one point, while no points are awarded for unsuccessful attempts.

The Global Articulation Test consists of thirty words and provides a detailed analysis of sounds, including both pathological sounds and those that meet the criterion for correct pronunciation. Correctly pronounced sounds are marked with a + (indicating normal pronunciation), while sounds that are softened (distorted) or irregular (where one sound is substituted for another) are marked with \pm . Sounds that do not appear in the word are marked with - (indicating omitted sounds).

The patient attended the speech therapy clinic twice a week for the first month, then once a week for the following five months. His daughter and wife were trained to assist him during daily activities.

During the treatment, we conducted oral practice exercises, voice articulation exercises using vowels in words and shorter sentences, as well as swallowing exercises to enhance the strength and mobility of the muscles essential for the swallowing process.

Outcome of applied treatment

Table 1 presents the results of the Water Swallowing Test and the Oral Practice Test conducted before speech therapy rehabilitation. In the water swallowing test which lasted 15 minutes, we got Profile 5 - the patient often chokes and it is difficult for him to drink all the water.

After six months of continuous speech therapy rehabilitation, the patient was re-evaluated using the Water Swallowing Test, which lasted five minutes. He achieved Profile 2, indicating that he can drink all the water in two or more sips without choking. The patient experienced no choking issues, as he continued to use the supraglottal swallowing technique.

On the 21-item Oral Practice Test, conducted before speech therapy rehabilitation, the subject scored three points for successfully completing three tasks, while he was unsuccessful in 18 tasks. Our results from the Oral Practice Test, which has not been administered to these patients previously, indicate weakness of the orofacial musculature. This reflects an inability to exert voluntary control, which is associated with impaired planning and programming of movements for speech, as well as the activities of chewing and swallowing food.

In the Oral Practice Test conducted after six months, the subject scored six points for successfully performing six tasks, while he was unsuccessful in 15 tasks. The results indicate minimal improvement, which did not significantly enhance the motor abilities of the oral structures, consistent with findings from other studies. With these exercises, the functionality of the movement of the speech organs is achieved.

In Table 2, the Global Articulation Test conducted before speech therapy rehabilitation indicated damage to all 30 phonemes, manifesting as distortion (9/±), substitution (15/±), and omission (6/-). This was particularly pronounced in spontaneous speech, making it difficult to understand. After six months, the patient distorted 10 phonemes (±), substituted 14 (±) and omitted 6 (-), indicating that there was no significant change in the patient's speech.

The ethical commission of the Sveti Vračevi Public Health Institution HOSPITAL in Bijeljina approved the study and consent was obtained from the subject himself and his family members. The research was conducted according to the Declaration of Helsinki.

DISCUSSION

We present a patient diagnosed with PSP, with pronounced swallowing and speech symptoms, who underwent speech therapy rehabilitation.

As the disease progresses, patients with progressive supranuclear palsy develop difficulties with swallowing and speech [12]. Dysphagia is the third most common symptom reported by PSP patients, i.e. patients report a swallowing disorder three times more often than a speech disorder [13]. Some authors note that dysphagia in PSP patients occurs approximately three to four years after the onset of symptoms [14], which aligns with our case study. In these patients, the oral/preparatory and oral phases of swallowing are most often impaired [15]. In our case study, the swallowing disorder was also manifested during the oral/preparatory and oral phases. During the Water Swallowing Test, which lasted 15 minutes, the patient received Profile 5 - indicating that he often choked, and it was difficult for him to drink all the water. The patient

experienced issues with bolus preparation and transport, attributed to weakened labial occlusion, and limited tongue mobility which are essential for initiating the voluntary aspect of swallowing and transporting food over the base of the tongue, resulting in frequent choking.

In the treatment of patients with dysphagia, both direct and medical procedures are applied. In this case study, we utilized direct procedures (including some of the exercises for the preparation and oral phase of swallowing) along with oral motor exercises. Additionally, we implemented a supraglottic swallowing maneuver to voluntarily protect the airway. The supraglottic swallowing maneuver is a technique that helps reduce or control aspiration during the oral phase of swallowing.

The treatment of these patients involves exercises aimed at improving the coordination of the muscles necessary for the swallowing process or stimulating the swallowing reflex. Additionally, postural maneuvers (such as turning the head and tucking the chin position) are employed to redirect the flow of the bolus, helping to protect the airways and facilitate swallowing. Then the patient is also advised to take small sips of water and bites of food, to cough and clear the throat after swallowing and to modify the consistency of the food (soft/mixed/pureed) [16]. However, as the disease progresses and patients become unable to swallow, some medical procedures are performed, most commonly the placement of a nasogastric tube or PEG (a surgical procedure known as percutaneous endoscopic gastrostomy) [17].

The orofacial musculature plays an important role in feeding, chewing, swallowing, speaking and facial expression. People with weakened oral musculature often experience difficulties with chewing/swallowing food as well as speech/language difficulties [18], which is consistent with our case study.

A basic prerequisite for the correct pronunciation of sounds is a well-developed oral practice. This means that the muscles involved in speech can perform all the movements necessary for their proper articulation as well as for the process of chewing/swallowing food. In the Oral Practice Test conducted after six months, the subject scored six points for successfully performing six tasks, while was unsuccessful in 15 tasks. The results indicate minimal improvement, which did not significantly enhance the motor abilities of the oral structures, in accordance with the research of other studies [19].

Good anatomy and mobility of the speech organs enable adequate speech. If one of these aspects is impaired to a lesser extent, it will have a negative impact on speech. Speech disorders in PSP are manifested by weakness, incoordination, paralysis or paresis of the speech muscles,

along with physiological characteristics that include disturbances in the speed, strength, order and accuracy of muscle movements [20], which is also characteristic of our case study. That is, the patient in our case study had dysarthria (slow, slurred and poorly articulated speech) [21], characterized by reduced mobility, coordination, and precision of the oral musculature.

With the global articulation test, we found impairment of all phonemes by distortion and omission type, which was particularly pronounced in spontaneous speech and made it more difficult to understand [22].

Good articulation is directly related to oral practice, meaning that the muscles involved in speech must perform well all the movements necessary for proper articulation. Speech rehabilitation in patients with PSP refers to exercises for the mobility of the speech organs, proper speech breathing, appropriate pace and rhythm of speech, and proper intonation of the voice. With these exercises, the functionality of the movement of the speech organs is achieved. According to the Global Articulation Test after conducted after six months, our patient's speech did not change significantly. Dysarthria remained present, along with impairments in a large number of phonemes due to distortion, substitution, and omission [22, 23], indicating the need for continued speech therapy rehabilitation.

Parkinsonism, vertical paralysis of the gaze, postural instability accompanied by frequent backward falls and cognitive impairment can significantly impair or shorten the life expectancy of these patients. Therefore, it is essential to provide increased support to both the patients and their family members in the future [24, 25].

The swallowing and speech rehabilitation program are among the procedures that should be recommended to patients at the very beginning of the onset of the above-mentioned symptoms.

Conflict of interest: None declared.

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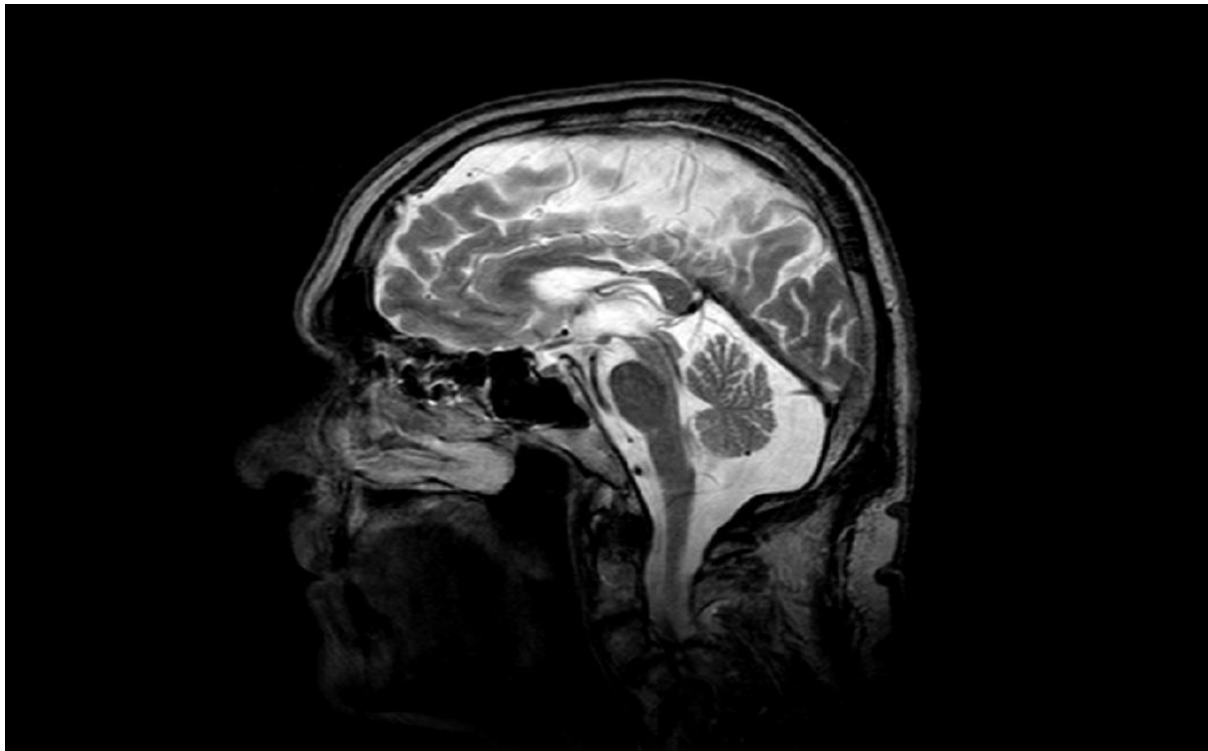


Figure 1. Magnetic resonance on the sagittal tomogram shows the so-called sign. hummingbird and discretely elevated tegmentum and tectum signal; a lower ratio of the diameters of the pons and midbrain is present, as one of the differentiation factors in relation to Parkinson's disease

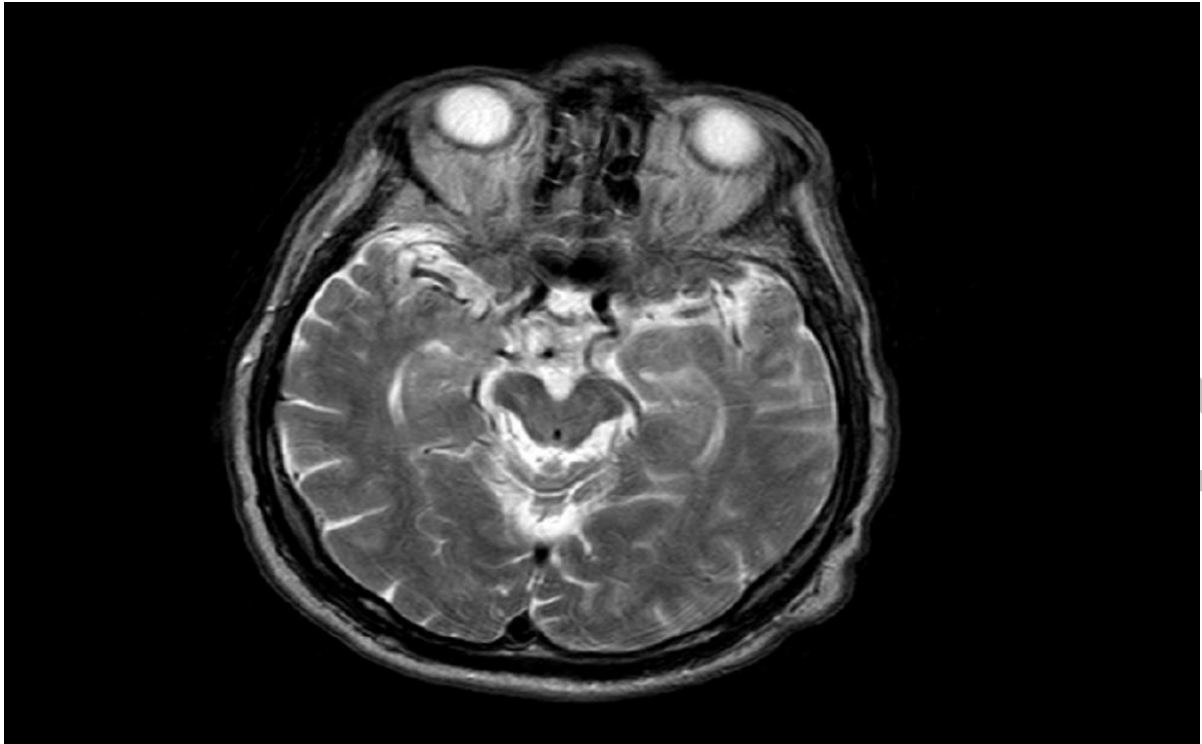


Figure 2. Magnetic resonance imaging shows significant atrophy with concave edges of the mesencephalon

Table 1. Results of the water swallowing test and oral practice before and after speech therapy

Test	Before therapy	After therapy
Water swallowing test	Profile 5	Profile 2
Oral practice test (task successfully completed / number of tasks)	3/21	6/21

Paper accepted

Table 2. Results of the global articulation test before and after therapy

Time	Distorted and substitution voices	Omitted voices
Before therapy	24/30	6/30
After therapy	24/30	6/30

Paper accepted