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A SPECIFIC PICTURE OF SPEECH DISTURBANCES IN POLISH SPEAKING PATIENTS WITH MIXED DYSARTHRIA IN MULTIPLE SCLEROSIS (MS) AND IN WILSON'S DISEASE (WD)

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SUMMARY

Background: The aim of this paper is to compare a specific picture of speech disturbances in Polish-speaking patients with mixed dysarthria in multiple sclerosis (MS), and in Wilson's disease (WD).

Material/ Methods: We selected for the experiment 20 persons with dysarthria of different origin: 10 persons with dysarthria in multiple sclerosis (MS), and 10 persons with dysarthria in Wilson's disease (WD). The compared groups were similar in age and the severity of the dysarthria. Patients read 11 words containing selected Polish sounds distinguished according to the manner of articulation. The results were qualitatively and quantitatively analyzed (using a non-parametric Mann-Whitney U test).

Results: The picture of speech production disturbances in Polish-speaking patients with mixed dysarthria in multiple sclerosis (MS), and in Wilson's disease (WD) includes: (1) the interpenetration of motor and linguistic problems, (2) variability in sound production, (3) the relationship between cognitive, emotional and motor speech functions, (4) production disturbances concerning one or more articulatory features of sounds. However, we found significantly more disturbances in the production of fricative sounds in the group with WD than in the group with MS.

Conclusions: The specific picture of speech production disturbances in Polish-speaking patients with mixed dysarthria of different origin is related to the etiology and pathomechanism of dysarthria. There is significantly more disturbances in the production of fricative sounds in the group with multiple sclerosis (MS), than in the group with Wilson's disease (WD).

Key words: motor speech disorders, fricative sounds

BACKGROUND

In the subject literature there is no example of extensive comparative research documenting the characteristics of speech in Polish-speaking dysarthric patients with various neurological conditions and the various types of dysarthria resulting from them (Pąchalska et al. 2014).

In view of the differences between language systems, it is not possible to transfer foreign research findings concerning the manner of speech sound production in patients speaking a foreign language (e.g. English) to Polish-speaking patients. Therefore, after making oneself familiar with the findings of long-term foreign research into dysarthrias (Murdoch 2010; Duffy 2005; Ala et al. 2011), it is necessary to conduct independent research on Polish-speaking people with dysarthria[1].

The lack of multifaceted research into the production disturbances observed in Polish-speaking dysarthric patients limits the possibility of developing complex principles of treatment based on a scientifically proven analysis of the pathomechanism of these speech disorders. On the one hand, it is true that patients with the same neurological damage may manifest different symptoms of speech disorders; on the other hand, some clinicians state that it is possible to detect aurally distinctive features of the speech of people with dysarthria resulting from a specific medical condition (Taly 2007; Murdoch 2010).

These opinions induced the authors of this article to look for those potentially distinctive features that could be reigistered register in people with various types of dysarthria. This is especially important in the case of mixed dysarthrias; something which has to date not been described explicitly within the specialist literature due to the complexity and diversity of disturbances as equally in patients with multiple sclerosis (MS), as for those in the group with Wilson's disease (WD).

Dysarthria is understood as a motor speech disorder (Murdoch B.E., 2010; Duffy J.R., 2005). According to this point of view, the phonetic aspect of an utterance (secondarily phonological aspect) is disturbed in dysarthria, while the lexical and grammatical aspects remain intact (see also Pachalska et al. 2014). The symptoms of production disturbances in individuals with dysarthria are usually analyzed in connection with the efficiency of the effectors of the articulatory organ. This position is justified as the paralyses and pareses that occur in dysarthric patients affect the muscles of the respiratory, phonatory and articulatory systems and result in speech production difficulties. However, this does not exhaust the issues in their entirety.

[1] It is worthy to note that dysarthria is probably the most common neurologic manifestation of Wilson's disease (Wilson 1912), found in 85 to 97% of those with neurologic Wilson's disease (Lorincz 2010). Dysarthria in Wilson's disease is most frequently of the mixed type with varying spastic, ataxic, hypokinetic, and dystonic components (Starosta-Rubinstein et al. 1987; Svetel et al. 2001). Speech involvement is frequently concordant with the neurologic involvement. In those with dystonia, speech frequently will have dystonic qualities with a strained or harsh quality. In those with parkinsonism, the speech quality may have hypokinetic properties. Ataxic dysarthria, with variation in word spacing and volume, is often found in association with other types of dysarthria, and may be more common in those with tremors (Starosta-Rubinstein et al. 1987).

Clinical observations made while working with people with dysarthria of various etiologies and the research findings presented in this paper induce a broader approach to the issues being discussed as they indicate that the emotional and intellectual functioning of people with dysarthria influences the way they produce speech. Such an approach to the issues being discussed is in conformity with the results of neuropsychological research which proves that the subcortical structures and the cerebellum are involved both in higher linguistic functions and in controlling verbal and motor functions (Pąchalska 2007; 2010; Pachalska et al. 2014). Since the cortex and the subcortical structures are functionally connected, patients with dysarthric disorders may have organic and reactive emotional disorders which influence the manner of speech production and the picture of production disturbances as well as the possibility of active participation in speech therapy. Taking into consideration these mechanisms is in conformity with the microgenetic approach of Pąchalska (2010). She presents the speech act as the final effect of the emotional and cognitive processes taking place within the speaker's psyche. This approach also justifies the comprehensive, multi-specialty treatment of people with dysarthrias[2].

In our study we adopted the definition of dysarthria by Darley (1975) and by Pachalska et al. (2014), according to which dysarthria is a group of respiratory, phonatory and articulatory dysfunctions resulting from damage to the motor part of the central and/or peripheral nervous system and disturbed muscular control over the speech production apparatus due to this damage. In our understanding dysarthria is divided into five basic types including **mixed dysarthria,** the topic of our research. We will perform analyses on the phonological system and the descriptions of normal productions provided by Rocławski (2001). The complexity and ambiguity of the symptoms of speech disturbances in dysarthrias of various etiologies, especially in mixed dysarthria, which occurs in both of the groups under comparison, was also undertaken for this comparative research[3].

The aim of this paper is to compare the specific picture of speech disturbances in Polish-speaking patients with mixed dysarthria in multiple sclerosis (MS), and in Wilson's disease (WD). The study is exploratory, based mainly on clinical observations, and analyzes the manner of sound production in word reading in patients with dysarthria secondary to multiple sclerosis (MS) and with dysarthria due to Wilson's disease.

Tools

A personal computer (Excel for calculations and Word for data entry).

[2] Pachalska (op. cit.) provided a detailed description of therapeutic interventions connected with different types of dysarthria in a separate publication. The scope of this paper does not allow for elaboration on this subject.
[3] According to the subject literature, Wilson's disease may involve mixed dysarthria with elements of rigidity, spasticity, ataxia and dyskinesia, while multiple sclerosis involves mixed dysarthria with elements of spasticity and ataxia. The author has provided a detailed description of the production disturbances observed in patients with Wilson's disease in the article Zaburzenia mowy w chorobie Wilsona [Speech disorders in Wilson's disease], published in Poradnik Jezykowy, 2007, no. 8, pp. 66-79.

MATERIAL AND METHOD

We selected for the experiment 20 persons with dysarthria of different origin: 10 persons with dysarthria in multiple sclerosis (MS), and 10 persons with dysarthria in Wilson's disease (WD). The compared groups were similar in age (see: Table 1) and the severity of the dysarthria.

Patients read 11 words containing selected vowels and consonants of the Polish language of each class distinguished according to the manner of articulation. **The production of the following words was analyzed and compared: DOM (house), GĘŚ (goose), JAR (ravine), LEW (lion), MASŁO (butter), NIEBO (sky), NOŻYCE (scissors), PAJAC (clown), POMIDOR (tomato), REGAŁ (bookshelf), SIEŃ (hall).** The indicator of the severity of production disturbances in dysarthria was the number of sounds in the material (words) produced in a disturbed manner. We completed and compared the disturbances in the articulation of those with dysarthria in multiple sclerosis (MS), with the disturbances in articulation experienced by those with dysarthria in Wilson's disease (WD).

Careful selection of the examined patients can guarantee that the results are unable to be falsified by the impact of such additional variables as gender and the disturbance severity level on the production of speech sounds in the groups under comparison.

Table 1. The characteristics of the examined groups.

Examined groups	Men	Woman	Total
Multiple Sclerosis (MS)	5 (50%) 44.6 (SD =1.4)	5 (50%) 43.9 (SD =1.4)	10 44.1 (SD =1.2)
Wilson's disease (WD)	5 (60%) 43.7 (SD =1.2)	5 (40%) 44.3 (SD =1.2)	10 44.2 (SD =3.1)
Total	10 (55%)	10 (40%)	20 (100%)

RESULTS

The data was analyzed in two stages:

1. In the first stage, the production disturbances in the group of people with multiple sclerosis (MS) were qualitatively described and compared to the qualitative results for phoneme production in those with Wilson's disease (WD), which had been obtained previously. A qualitative analysis was conducted. This type of analysis consisted of:
   a. Comparing sound productions in the participants of both groups with productions that are correct in terms of orthophony given in the relevant subject literature in order to determine deviations from normal production.
   b. Describing the approximate position of the articulators during the production of sounds found in the material that was read out, as well as disturbances in their production.

c. Determining the disturbed articulatory features observed in each of the groups concerned.

2. In the second stage, the results were quantitatively analyzed. The quantitative analysis consisted of:
   a. determining the number of disturbed productions in each class of sounds in both groups.
   b. comparing the groups in terms of the statistical significance of the differences in the severity of production disturbances in the individual classes of sounds depending on the etiology of the dysarthria by using the non-parametric Mann-Whitney U test.

Qualitative analysis

The qualitative analysis showed disturbances in the production of sounds for all the distinguished classes (Table 2). Voicing problems occurred in the production of plosives (partial devoicing and complete devoicing in the group of people with multiple sclerosis, and partial devoicing in the group of people with Wilson's disease) (Table 3).

Also, simultaneous problems with voicing and oral realization manifested themselves in the group of patients with multiple sclerosis ([p] was replaced with [m]). This difficulty was related to the symptoms of ataxia, which was the pathomechanism of the patient's difficulties in coordinating the phonatory apparatus and the soft palate. The alveolar production of [d] in the word "*dom*" [house] in a patient with Wilson's disease might have resulted from his ataxia, which causes difficulties in making a precise movement and "hitting" the place of articulation.

The production of fricatives was distorted significantly more frequently in the group of people with Wilson's disease than in the group of people with multiple sclerosis, which manifested itself in difficulties with maintaining articulatory positions or changing the shape of slots so that the fricative production may have resulted from the patients' dyskinesias (tremors) and increased muscle tone, which made it impossible to acquire the appropriate shape of the slots. The bilabial production of [f], which resulted from the pathologically increased tone in

Table 2. Total number of disturbed sound productions in each class of sounds in the group of patients with multiple sclerosis (MS)

Patients with multiple sclerosis (MS)							
Disturbed sound	**Plosives**	**Fricatives**	**Affricates**	**Sonorants**	**Approxi-mants**	**Vowels**	**Total number of disturbed produc-tions**
Total number of disturbed sound productions	12	4	10	26	2	24	**78**
MEAN	1.20	0.40	1.00	2.60	0.20	2.40	**7.80**
SD	1.55	0.52	0.75	1.17	0.41	4.43	**6.50**

Table 3. Total number of disturbed sound productions in each class of sounds in the group with Wilson's disease (WD)

Patients with Wilson's disease (WD							
Disturbed sound	**Plosives**	**Fricatives**	**Affricates**	**Sonorants**	**Appro-ximants**	**Vowels**	**Total number of disturbed produc-tions**
Total number of disturbed sound produc-tions	4	28	8	18	2	12	**72**
MEAN	0.40	2.80	0.80	1.80	0.20	1.20	**7.20**
SD	0.55	1.30	0.84	2.17	0.45	0.84	**4.97**

the orbicularis oris muscle and dystonia in the form of a grimace of a smile, making it difficult to form a slot accurately; this was found only in the group with Wilson's disease. In the group of people with multiple sclerosis, distorted production of fricatives was noted only once, taking the form of an interdental production of [s].

Both groups had difficulty in achieving closure, which resulted in an abnormal production of affricates (e.g., the production of [ʦ] with incomplete closure and a rustling sound resembling [s]). These difficulties turned out to be more increased in those with multiple sclerosis than in the patients with Wilson's disease. In the group of patients with multiple sclerosis, the stop - fricative sequence was broken up in the production of the affricate [ʦ], which may have resulted from the symptoms of spasticity (the patient who manifested these symptoms had a very high muscle tone, distinctly limiting and slowing down his tongue movements). This pathology resulted in the production of two sounds: [t] and [s]. Also in the group of patients with multiple sclerosis, a complete lack of closure was found, resulting in the production of [s] instead of [ʦ]. This manner of production was related to the weakened tongue muscles due to increased tone.

The production of sonorants was disturbed in both groups; however, there were differences in the quality of the disturbances. In the group of patients with Wilson's disease, dorsal production of [l] was found as well as the production of [l] with a tense, narrowed tongue, with the apex raised to the upper teeth, and distorted production of [ɲ] as a nasal spirant sound without moving the mass of the tongue forward, which resulted from an increased tone in the tongue and its rigidity. Such positions of the tongue were not observed in patients with multiple sclerosis.

In the group of people with multiple sclerosis, spasticity resulted rather in a tendency to produce slower and more limited movements, which brought about, for example, the production of [ɲ] as a palatal nasal component [n̆] and the [j] sound, and a flap [r].

In the group with multiple sclerosis, the abnormal production of [j] was found, taking the form of inaccurate production, with a lowered tongue position resulting in a sound similar to [ɨ].

As far as vowel production is concerned, a nasalization of oral vowels was found in both groups. Also, in the group of people with Wilson's disease there occurred the two-phoneme production of the diphthong ɛ + nasal labial component [ɛ̃ʷ]/ɛ + nasal palatal component [ɛ̃ʲ] in the word "*gęś*" [goose], which was produced as [gewɛ]. Such two-phoneme production may have resulted from slow movements and the pathological prolongation of the production of the dynamic nasal vowel. A larger number of non-systemic productions in the form of sounds intermediate between one vowel and the other occurred in the patients with Wilson's disease, while the nasalization of oral vowels occurred in those with multiple sclerosis more frequently.

In the patients with multiple sclerosis, respiratory-phonatory incoordination manifested itself in difficulty with exhalation control and in voice weakening, which was reflected in a larger number of disturbances concerning voicing. In the group with Wilson's disease, the symptoms of incoordination manifested themselves in two patients with ataxia symptoms – they consisted of abnormal placement of the breathing pauses.

Prosodic disturbances were found both in the patients with Wilson's disease and those with multiple sclerosis; scansion, drawling and additional vocalic elements dominated in the patients with Wilson's disease while a shortening of sounds was found less frequently in this group. Different durations of sound productions were more frequent in the group of patients with multiple sclerosis.

Not only did the participants' motor efficiency but also their emotional state influence their manner of speaking. A markedly low mood was to occur more frequently in the patients with multiple sclerosis. Some patients experienced direct emotional difficulty in confronting their evident speech disorders (statements such as: "I'd be glad to talk to you, but don't examine me, please"). Also slight cognitive difficulties were noted in the group of patients with multiple sclerosis (two patients experienced difficulty in focusing and remembering instructions). It was not verified whether these difficulties were secondary to the patients' mood disorders or if they resulted from primary cognitive dysfunctions. Such difficulties did not occur in the group of patients with Wilson's disease despite a similar severity in the speech disorders.[4]

Variability in the production of sounds occurred in both groups. The participants with reduced speech intelligibility did not always distort the sounds in the same manner (e.g., both the flap [r] and the productions of [r] with the slot instead

[4] In a group of people with Wilson's disease with a different level of production disturbances than in the group of patients with multiple sclerosis - not included in the quoted studies - emotional and cognitive difficulties manifested themselves in the form of an incomplete insight into the deficits experienced. In addition, some patients reported that the selecting of the appropriate word difficulties were one of the first speech disturbances preceding the occurrence of dysarthria.

Table 4. Number of production disturbances in the individual classes of sounds in both groups – Statistical analysis

Sound production in words					
Number of disturbed productions	**Patients with Wilson's disease (WD) N = 5**		**Patients with multiple sclerosis (MS) N = 5**		**Mann-Whitney U test p=0.05**
	Mean	**SD**	**Mean**	**SD**	
Plosives **TD**	0.40	0.55	1.2	1.55	12.5
Fricatives **SZ**	**2.80**	**1.30**	**0.4**	**0.52**	**1**
Affricates **TsDz**	0.80	0.84	1	0.75	10.5
Sonorants **L**	1.80	2.17	2.6	1.17	8.5
Sounds [as above] **W**	0.20	0.45	0.2	0.41	9
Vowels **V**	1.20	0.84	2.40	4.43	8.5

Table 5. Number of disturbances in articulatory features in the group with Wilson's disease (WD)

Patients with Wilson's disease (WD)					
Articulatory features	**Sonority**	**Nasality**	**Place of articulation**	**Stricture**	**Position of the tongue body**
Number of disturbances in articulatory features	**6**	**4**	**46**	**40**	**8**
MEAN	**0.60**	**0.40**	**4.60**	**4.00**	**0.80**
SD	**1.34**	**0.89**	**4.16**	**1.22**	**1.30**

of the vibration of the apex without vibrations were found in the same person with multiple sclerosis). In the patients with Wilson's disease with increased muscle tone, the extent of variability was smaller although variable productions did also occur.

The participants' speech intelligibility was reduced to a varying extent – in the group of people with Wilson's disease, reduced intelligibility was found in three patients. A similar result concerning speech intelligibility problems was found in the group of people with multiple sclerosis. Non-systemic productions and disturbances in the articulatory features of the sounds produced occurred in these patients, making phonological identification difficult (e.g., productions intermediate between [ɕ] and [ʃ]). At the same time, the reduced intelligibility of utterances was proof of the interpenetration of motor and linguistic disturbances – having a secondary effect on the phonological aspect of the text, deviations in the phonetic structure of an utterance changed the listener's possibility of receiving the content.

Significant individual differences in the state of speech in terms of the type and number of disturbances were observed in both groups.

Quantitative analysis

Quantitative analysis of production disturbances in the individual classes of sounds[5]

There were significantly more disturbances in fricative production in the group of people with Wilson's disease than in the group with multiple sclerosis.

There were more production disturbances in the remaining classes of sounds in the group of people with multiple sclerosis with a similar overall severity of dysarthria, but the differences between the groups being compared were not statistically significant due to individual differences in the state of the participants' speech and the scatter of results.

Quantitative analysis of disturbances in the individual articulatory features.

The differences in the number of disturbances concerning the individual articulatory features were not statistically significant in the groups compared.

DISCUSSION

The study confirms the significantly individualized nature of the state of speech in dysarthria of a similar etiology. It is a well know fact that there are differences in the manner of speech production between patients with different neurological conditions and the dysarthria of different etiologies (the significant difference in the severity of disturbances in fricative production) (Pachalska et al. 2014).

It was found that the speech of the participants with dysarthria secondary to Wilson's disease is marked by a significantly larger number of disturbances in the production of fricatives than the speech of the patients with dysarthria in multiple sclerosis. Such a picture of disturbances results from the pathomechanism of dysarthria in this disease (the dyskinesias that occur in the participants make it difficult to maintain articulatory positions, while increased muscle tone makes it difficult to achieve the appropriate shape of the slot).

The number of production disturbances was larger in the group of patients with multiple sclerosis than in the groups of patients with Wilson's Disease (WD) in most of the classes of sounds; however, the difference between the two groups (concerning the number of disturbed productions in the individual classes of sounds) was not statistically significant.

The picture of production disturbances might be complex – motor and linguistic problems are observed to interpenetrate.

Disturbances of speech production in people with dysarthria of various etiologies may concern one or more articulatory features of a sound and not every disturbed production results in reduced speech intelligibility. Reduced intelligibility occurs when the articulatory feature that determines the phonological identification is disturbed.

[5] Detailed results concerning the number of disturbances in the individual classes and the results of statistical analyses are provided in the tables at the end of the article.

The manner of sound production in people with dysarthria might be variable to a greater or lesser degree. Short-term production variability in dysarthrias depends on the type of dysarthria (variability is greater in those with ataxic dysarthria and hyperkinetic dysarthria than in individuals with increased muscle tone), the level of muscle fatigue in the speech production apparatus, the general level of arousal and the emotional state in patients (see also Pachalska 2010).

The borderline between production disturbances and prosodic disturbances in dysarthrias is fluid (e.g., the slower rate of tongue movements results in the breaking-up of the stop - fricative sequence in affricate production, and elisions and sound repetitions result in the change of the speech rhythm and rate).

When the manner of speech sound production in dysarthric patients is analyzed, it is necessary to remember that motor activities connected with speaking are the final stage of the emotional and cognitive processes connected with transmitting the content and "devising utterances" and they cannot be examined separately from the previous stages.

This comparative study is preliminary in nature. On account of the small samples analyzed and the limited language material as well as the considerable individual differences between the patients, it is necessary to be prudent in generalizing the findings. In spite of the above limitations, the study contributes new information to the Polish literature on speech therapy and if its findings are interpreted with prudence, it could constitute a guideline in speech-therapy assessment for patients with mixed dysarthria secondary to Wilson's disease and multiple sclerosis.

Clinical implications resulting from the study

It is necessary to include both the quantitative analysis and qualitative analysis[6] of specialist research findings (including speech-therapy research findings) in a speech-therapy assessment for patients with dysarthrias of various etiologies. **It is not possible to give the picture of the state of these patients' speech only on the basis of a statistical analysis of the number of disturbances in sound production.**

The assessment of the neuromotor pathomechanism of the symptoms of speech disorders is the point of departure for the motor therapy of speech functions. Such an assessment involves analysis of a patient's motor abilities. The therapist should trace the dynamics of disturbed productions (the observation of how the patient's tongue, lips and jaw work when he or she is speaking and the position of his or her head and trunk).[7]

[6] The use of objective and subjective assessment tools would be of considerable importance for gaining a detailed insight into motor abilities.

[7] In the light of the production disturbances observed, the traditional division into substitutions and distortions seems insufficient – an understanding of the pathomechanism and a qualitative description of production disturbances are of great significance, which is emphasized also by J. Panasiuk and D. Pluta-Wojciechowska in their publications.

The qualitative analysis of the research findings shows that the improvement of patients' cognitive and emotional functioning is often an important factor that makes an impact on speech therapy effects. For this reason, it is necessary to provide multi-specialist support for people with dysarthria that includes medical, physiotherapeutic, psychological and neuropsychological interventions (see also: Burke et al. 2011). The relationship between the pathomechanism of disturbances and the state of patients' speech justifies the need for adapting therapeutic interventions to the type of dysarthria and its severity. Thus, an integrated approach to the improvement of the motor efficiency of the speech production apparatus is recommended, which involves providing conditions for patients that will promote the optimization of muscle tone in the speech production apparatus (providing patients with psychophysical comfort during motor exercises).

Being aware of the relationship between the symptoms of disturbances and their multi-factor pathomechanism, the speech therapist is faced with new tasks. Speech therapists working with dysarthric patients should take into consideration not only their patients' motor deficits but the entire picture of their emotional and cognitive functioning (negative and positive diagnosis).

Bio-psychosocial determinants need to be taken into consideration while improving the functioning of patients with dysarthria if their motor speech system is to be efficiently improved (Brown & Pachalska 2003).

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