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Changes in temporal features of speech in secondary progressive multiple sclerosis (SPMS) – case studies

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ABSTRACT

Multiple sclerosis (MS) is a chronic inflammatory disease which, in addition to affecting motor and cognitive functions, may involve language disorders. Despite the importance of speech and language disorders in the quality of life of patients, there are only a few studies about language and speech production difficulties in MS. The aim of this research is to describe the limitation patterns of speech and temporal characteristics of the suprasegmental level in two SPMS cases related to various types of spontaneous speech tasks. We assumed the change of the cognitive load has a greater effect on spontaneous speech in MS patients than in controls. Two SPMS patients, and two sex-, age- and education matched healthy controls were studied. We applied verbal fluency tests (phonemic, episodic, semantic, verb), digit span test, non-word repetition test, Corsi Block Tapping Test, Stroop Colour and Word Test, and Trail Making Test. Token Test was used to measure speech comprehension. The four speech tasks required relatively different degrees of cognitive effort: (a) spontaneous narrative about own life; (b) event description; (c) picture description; (d) narrative recall. Our results show that there are differences between MS patients and controls: MS patients produced slower speech and articulation rate, and they had more and longer pauses in every speech task. Speech tasks and the degree of the cognitive load had a greater effect on MS patients than on control speakers.

Introduction

Multiple sclerosis is a chronic inflammatory disease of the central nervous system, one of the most common neurodegenerative diseases which affects sensory, motor and cognitive functions and may involve language disorders. Despite its broad occurrence, there are only a few studies about the speech and language functions in MS (review: Renauld, Mohamed-Said, & Macoir, 2016).

The majority of the cases are grouped in three different clinical courses: primary progressive MS (PPMS), relapsing-remitting MS (RRMS), and secondary progressive MS (SPMS), which develops from RRMS. The transition from RRMS to SPMS is a long-term process, its diagnosis is usually set up retrospectively (Lorscheider & Buzzard et al., 2016).

Studies have shown that in the progressive types of MS patients achieve lower scores in most cognitive and linguistic tests than RRMS patients (Chiaravalloti & DeLuca, 2008; Friend et al., 1999; Geisseler et al., 2016; Guimaraes & José, 2012; Johnen et al., 2017; Nocentini et al., 2001). SPMS patients are frequently reported severe symptoms in motor, sensory or cognitive areas. This type of MS develops gradually, in decades, since the first diagnosis, and the process of demyelination extensively affects white and gray matter areas.

Speech and language processes are frequently impaired (approx. 60%), but relatively less studied in MS (Renauld et al., 2016). Due to the different neurological status and the progression of the disease, language symptoms show great diversity. Many patients report some inconveniencies in their daily communication routine due to their articulation deficits, word finding difficulties, or other language symptoms. Although speech and language processes are strongly related to other cognitive processes, their relation and interaction in MS are relatively less studied (Kujala et al. 1996; DeLooze et al., 2017; Feenaugthy, Tjaden, Benedict, & Weinstock-Guttman, 2013; Yorkston et al., 2003). The communicative processes and speech tasks require complex reasoning, the ability of flexible attentional switching, complex memory processes, and fast real-time information processing.

Cognitive limitations in MS

Cognitive functions are affected in 65% of MS cases, especially the executive functions including memory, attention, and information processing speed (Rao, 2004). The deficits in cognitive domains cause greater difficulties in daily life for some patients than motor or sensory impairments (Nocentini et al., 2001). The limitations of some cognitive abilities are not evident or apparent in many cases, they can be concealed by other – primary – symptoms such as chronic fatigue or depression (Guimaraes & José, 2012). Chronic fatigue affects 90% of MS patients, depression affects 50% of them (Arnett & Stober, 2011).

Both short-term and long-term memory processes show more or less limitations particularly in the progressive types of MS (Connick, Chandran, & Bak, 2013; Piras et al., 2003; Rao, 2004). Numerous studies reported that verbal fluency tasks are the most sensitive to the detection of cognitive limitations in MS and strongly correlate with other verbal and speech limitations (Arnett & Stober, 2011; Beatty, 2002; Friend et al., 1999; Geisseler et al., 2016; Henry & Beatty, 2006; Kraus et al., 2005).

Attention deficits affect approximately half of the MS patients (Feenaugthy et al., 2013). These deficits are related to the delay of the information processing as well as to chronic fatigue (Arnett & Stober, 2011; Chiaravalloti & DeLuca, 2008; Rao, 2004). The decline of the information processing speed is shown particularly in progressive types of MS (Arnett, Smith, Barwick, Benedict, & Ahlstrom, 2008; Connick et al., 2013; Guimaraes & José, 2012; Rao, 2004), but – on a lower level – observed also in RRMS (Chiaravalloti & DeLuca, 2008). Patients have more difficulties in complex or time-limited tasks because they require more cognitive load (Arnett & Stober, 2011; Connick et al. 2013). The slowdown of information processing often has a negative impact on everyday activities (Chiaravalloti & DeLuca, 2008).

Narrative abilities in MS

Several studies show that narrative linguistic processes are affected by neurodegenerative diseases (e.g., Cuetos, Arango-Lasprilla, Uribe, Valencia, & Lopera, 2007; Forbes-McKay & Venneri, 2005; Gola et al., 2015; Hier, Hagenlocker, & Shindler, 1985; Thomas, Billon, & Hazif-Thomas, 2018). However, there are only a few studies which deal with conversational and narrative abilities appearing especially in MS (Arrondo, Sepulcre, Duque, Toledo, & Villoslada, 2009). Analysis of spontaneous speech might be useful means for the examination of background cognitive mechanisms, since both the temporal features of speech and syntactic and semantic content are sensitive to the difficulties in attentional and memory processes and processing of information.

Specific spontaneous speech tasks require the activation of different cognitive processes. During the production of narratives, episodic and semantic memory and working memory are being activated. In addition, the appropriate functioning of several other cognitive processes, like planning and decision-making processes, monitoring the environment, ignoring irrelevant stimuli, temporal sequencing, constructing of mental spaces and events, theory of mind, or organizing feelings, are necessary (Hirsch, Mar, & Peterson, 2013; Spreng, Mar, & Kim, 2008). Fewer cognitive resources might make the retrieval from specific memory systems more difficult (Donix et al., 2010), the results of which can be detected in several linguistic processes.

There are differences between the productions of narratives according to their topics (personal or non-personal) or their time span (past or future) (Abraham, Schubotz, & von Cramon, 2008). Remembering past events or future prospections activate different memory subsystems and different brain areas, and require specific constructive processes. According to Abraham et al. (2008) personal event description requires the appropriate functioning of autobiographical memory, episodic memory, as well as mental scene construction and re-construction ability, ability of constructive simulation, action planning and self-processing. According to Donix et al. (2010) the quality of the retrieval from the autobiographic memory might be predictive in the early stages of Alzheimer's disease. According to Gola et al. (2015) certain neurodegenerative diseases might be recognizable by analyzing spontaneous personal storytelling. They found that depending on the involved brain areas, there are typical changes in important functions for storytelling such as temporal organization, narrative evaluations, and social attention. Results show that characteristics of conversational storytelling are associated with the speech of processing and mental flexibility, while certain differences might be noticed during vocal analysis. Arrondo et al. (2009) found lower word count, shorter sentence length, and more function words in personal conversational narratives of MS patients.

Speech of patients with neurodegenerative diseases show typical differences from control speech in guided narratives (in non-spontaneous conversational situations) like picture describing and narrative recall often used in the assessment of acquired language disorders. Picture description requires a complex integration of communication capabilities, so it might be a sensitive – diagnostic – tool for detecting linguistic changes in mild cognitive impairment, Alzheimer's disease or dementia (Choi, 2009; Cuetos et al., 2007; Forbes-McKay & Venneri, 2005; Hier et al., 1985). Analyzing picture descriptions, differences were found between patients and healthy control speakers in the total number of words, number of unique words, phrase lengths, number of subordinate closes (Hier et al.,

1985), simplifying grammatical structures, decreasing information content, vocabulary, and word-retrieval strategies (Forbes-McKay & Venneri, 2005), fluency, and redundancies and number of empty words (Cuetos et al., 2007). During narrative recall, working memory, information processing speed, and attentional processes can be tested. This task is one of the most difficult speech tasks for both healthy young and old speakers (Bóna, 2014). Analyzing abilities of narrative recall of patients with mild cognitive impairment (MCI) and Alzheimer's disease, Park, Park, Sohn, Kim, and Park (2016) found that both groups show differences in the quality of recall compared to healthy controls, but the performances of the AD group were lower than those of the MCI group. This indicates that global cognitive functions are strongly correlated with the state of verbal memory. Goldstein et al. (1992) analyzed gist recall in MS patients. They found that narrative recall is a dynamic complex task in which the aim is not only to memorize the elements of the story, but to organize them into integrated narratives, activate elements of knowledge of the world to understand the story, create the proper logical structure, and ignore details irrelevant to the essence of the story. They found that the way in which the participants use the content of the narrative gives an idea of the effectiveness of semantic processing in their minds. The results show that the ability of gist recall is generally preserved in MS. In addition to examining semantic content in the narrative recall, observing temporal characteristics of speech is still an undiscovered area.

In this study, four narrative tasks were recorded with MS patients and control speakers. Two tasks were personal and two tasks were non-personal: spontaneous personal narrative, personal event/scenario description, non-personal picture description, and non-personal narrative recall. In the analysis, the changes in the temporal features in the narratives and the deviations from the control persons were analyzed.

Temporal features of speech in MS

In this paper, we examine the changes in temporal features of spontaneous speech, particularly articulation rate, speech rate, and frequency, duration and types of pauses. Similar changes in temporal features of speech have been observed in other neurodegenerative diseases, e.g., mild cognitive impairment (Tóth et al. 2018), Alzheimer's disease (Hoffmann et al., 2010; Meilán et al., 2014; Weiner, Neubecker, Mary, & Hynan, 2008), Parkinson's disease (Rektorova et al., 2016; Tjaden & Wilding, 2011), and Huntington's disease (Azambuja et al., 2012). Temporal characteristics of speech in MS are less described issues (DeLooze et al., 2017; Feenaugthy et al., 2013; Rodgers, Tjaden, Feenaughty, Weinstock-Guttman, & Benedict, 2013; Tjaden & Wilding, 2011).

The higher cognitive and linguistic demand and the time-limited or time-measured tasks require higher mental effort which might cause slower speech rate as well as more and longer silent and filled pauses (Arnett et al., 2008; DeLooze et al., 2017; Swets, Jacovina, & Gerrig, 2013). DeLooze et al. (2017) found that MS patients with cognitive impairments produced more dysfluencies, slower speech rate, more silent pauses compared to MS patients without cognitive impairments. Feenaugthy et al. (2013) measured slower speech rate and faster articulation rate, longer pauses and increased pause frequency both in cognitively impaired and non-impaired MS patients. They found higher effects of cognitive-linguistic demand on the low-performance MS-group than on the normal-performance MS-group. Tjaden and Wilding (2011) observed an increased amount of syntactically inappropriate pauses compared to the healthy control group.

Rodgers et al. (2013) detected differences between the speech rate of spontaneous speech and reading aloud. Their results support the hypothesis that the cognitive demand of the speech task has a greater effect on the changes in temporal features of speech in MS patients than in healthy controls.

Objectives and hypotheses

In this study, two MS patients with a similar clinical background and medical status were examined. We compared their cognitive tests and temporal characteristics of their speech. The speech tasks of the participants were to produce spontaneous and semi-structured narratives. Speech tasks were selected so that different cognitive resources were involved in the production. Our hypotheses were the following:

(H1) The speech task affects the temporal features of speech of patients and controls: involvement of different cognitive resources and relative difficulties of speech tasks manifest in decreased speech rate and articulation rate, and increased pause duration and frequency.

(H2) Due to cognitive difficulties MS patients are more affected by the increased use of cognitive resources than controls. We supposed that differences between the certain tasks will be more evident in MS patients than in controls.

(H3) Temporal features will be different between personal and non-personal speech tasks in each participant. Personal speech tasks are easier than non-personal tasks, and this will appear in the differences of speech rate and articulation rate. Difference between speech tasks will be higher in the speech of MS patients than in the speech of control speakers.

Methods

Participants

Two patients with SPMS and two healthy control speakers participated in the examination (Table 1). Each participant was a native Hungarian speaker with intact hearing, and no psychiatric or addiction history. Controls had no mental disorder, depression or neurological disease. Patients had normal speech comprehension ability which was measured by Token test (DeRenzi, 1962; scores: P1 = 35/37, P2 = 35/37). None of the patients were diagnosed with dysarthria. Neither before nor during the recording period did they attend speech and language therapy. There is no information available on medication and vision problems, but they took

Table 1. Participants (P: patients, C: controls).

No.	Sex	Age	Education (years)	First diagnosis (post onset, years)	Changes to SPMS (post onset, year)
P1	M	45	24	27	15
P2	F	56	16	25	12
C1	M	44	20	–	–
C2	F	53	17	–	–

their usual medications at the time of the examination. The EDSS (Expanded Disability Status Scale) score determined by their physician was also shown (P1: 8,5; P2: 6,0).

Neuropsychological tests

Complex working memory was assessed with standard tests: the phonological loop with digit span and non-word repetition test (Hungarian adaptation: Racsmány, Lukács, Németh, & Pléh, 2005); the visuospatial sketchpad with Corsi Block Tapping Test (Kessels, van Zandvoort, Postman, Kapelle, & de Hand, 2000), and the central executive with verbal fluency tasks including phonemic (letter "s"), semantic (category, animals), episodic (things available in a grocery store) and action (verbs) fluency. The time limit was 1 min for each fluency task. Proportion of pauses in the total speaking time of verbal fluency tasks was also measured. For measuring attention control and cognitive flexibility the Stroop Color and Word test were applied.

Material

Spontaneous speech was recorded in four speech tasks. Personal and non-personal tasks were used. Personal: (a) spontaneous narrative about own life; (b) describing an event of the subjects' own life; Non-personal: (c) picture description; (d) narrative recall (historical anecdote). Speech tasks were selected so that they required the involvement of various cognitive resources and subskills. We assumed that certain tasks would be easier, while others would be more difficult for patients to perform depending on the available resources. We also assumed that the relative difficulty level of each task will also be reflected in the speech parameters.

(a) In spontaneous narratives, subjects were asked to speak about their families, work or hobbies. They could speak freely, they had time to speak and they had a new question only when they could not continue their speech. In this task speakers could select their own ideas, linguistic structures, and expressions.

(b) The task was describing an event of the subjects' own life (scenario description task). In this study subjects were asked to speak about the family customs of Christmas celebrations. Since speech samples were recorded in summer and early autumn, subjects had to remember a more distant, special event, and tell it in chronological order.

(c) In the picture description task, subjects had to speak about a picture of an interior as elaborately as possible. There was a dining room and a kitchen in the picture, but some of the other rooms were partially visible.

(d) In the narrative recall task, participants listened to a 2-min-long unknown historical anecdote. Their task was to summarize the heard story as accurately as possible.

Speech samples were recorded digitally in a soundproof room with the cognitive examinations on the same day. The speech signal was recorded with an omnidirectional condenser microphone at 44.1 kHz sampling rate. One seating took about 1 h. Speech samples were elicited in the same order for both speakers: from the easiest to the most difficult. Between two speech tasks, there was a cognitive examination to avoid the

monotony of the tasks. From each speaker, altogether 10 min of speech were analyzed – the same length of speech samples (ca. 2.5 min per task) were examined from each speaker and each speech task.

Analysis

Annotation (utterances between two pauses) was made manually by Praat 5.0 (Boersma & Weenink, 2008) in the speech samples (spontaneous narratives, scenario description task, picture description task, narrative recall). Duration of silent and filled pauses and utterances was measured automatically by a Praat script. Speech rate (the total number of sounds divided by total speaking time including pauses) and articulation rate (the total number of sounds divided by total speaking time without pauses), the proportion of pauses in the total speaking time, mean duration of speech units, frequency of silent pauses and filled pauses were calculated for all speakers. The speech rate and articulation rate were calculated in sounds/sec (instead of syllables/sec), so that the data can be used in the later automatic analysis. The measures were based on the number of realized units, and not on the numbers of "intended" units (Koreman, 2006). The data were compared across the speakers and the four speech tasks.

Ten percent of the measurements and all calculations were carried out twice by the second author, 2 weeks apart. The occurrences of pauses and their boundaries were compared in the two analyses. If there was only 10 ms difference between the annotations of the pause boundaries, they were considered the same. The results of the two analyses were similar in 100% of the cases.

Results

Neuropsychological tests

The results of the neuropsychological tests are shown in Table 2. Digit span and non-word repetition tests were evaluated according to Hungarian standard scores (Racsmány et al., 2005). Corsi Block Tapping test was rated by international standards (Kessels et al., 2000). Hungarian data under standardization were used for the phonemic, semantic and episodic fluency results (Tánczos, 2014). Hungarian standard for action fluency was not available; therefore, we used Woods et al. (2005) data for measuring. In fluency tasks, the results of the patients and controls are shown in Table 2. Comparison with standards is shown in

Table 2. Results of the neuropsychological tests.

Test	P1	P2	Reference value
Digit span test (item)	5	4	6,85 (SD: 0,67)
Non-word repetition test (item)	5	5	5, 85 (SD: 1,22)
Corsi block tapping test (item)	4	4	6,2 (SD: 1,3)
Verbal fluency (word)			
Phonemic	19	15	18 (range: 17 − 19)
Semantic	26	11	28 (range: 25 − 31)
Episodic	20	14	26 (range: 23 − 29)
Action	29	Nouns instead of verbs (11 word)	36,5 (range: 34 − 39)
Stroop test (%)			
I. baseline	Baseline	Baseline	Baseline
II. color	I.+100	I.+71	I.+17
III. color + word	I.+423	I.+286	I.+42

Table 3. Results of the verbal fluency tasks (percentile).

(percentile)	P1	P2	C1	C2
Phonemic fluency	50	75	50	75
Semantic fluency	50	10	50	75
Episodic fluency	25	25	75	75
Action fluency	90	10	100	100

Table 3. Stroop test was compared between patients and controls. For tests where valid standard Hungarian normative data (digit span task, non-word repetition task) are available or measurement is language-independent, international standards (Corsi Block Tapping Test) can be used. In these cases, patient's data were compared to normative data. This allowed a more accurate comparison than the comparison with two controls. For tests where standardized or non-standardized data were available (verbal fluency task, Stroop task), the task was recorded with the patients and the control persons for comparability.

Patients had significantly lower performance in the digit span task than that of the standard score, their scores fell >2SD below standards. Both patients achieved a normal score in non-word repetition task (<1SD). Their performance fell >1,5SD below the standard scores in Corsi Block Tapping Test. Greater difference was found between Stroop test I., II. and III. in patients than in controls. This means that the overall duration of response time and task performance of patients in Stroop II and III tasks increased significantly compared to I. P2 had a higher performance in Stroop test than P1. In the performance of P1, there was a tenfold difference compared to controls in Stroop II. and III. Fourfold (in Stroop II) and approximately sevenfold slowing (in Stroop III.) were shown in the performance of P2 compared to controls.
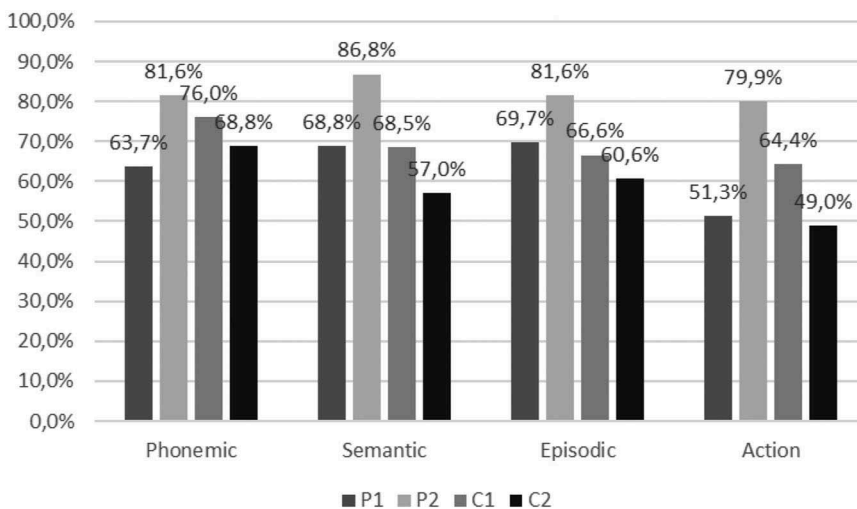
Results of verbal fluency tasks did not show a consistent tendency. Results are presented in percentile because of different standards (Table 3). Control persons achieved at least average performance in each task. P1 had higher performance in each fluency task relative to P2.

Both patients completed the phonemic fluency task similar to controls. In semantic fluency task, P1 achieved a similar score as controls, P2 performed quite below average. Patients had a lower, at the bottom of the average score in the episodic task relative to controls. The words in the action fluency task were scored in the performance of P2, despite the use of noun forms of actions instead of verbs (she said *futás* [noun, *running*] instead of *fut* [verb, *to run*]). The performance of P2 was also extremely low in this task. In action fluency task, P1 achieved above-average score, similar to controls. As in the speech tasks (see Table 4), we measured the proportion of pauses in the total speaking time of verbal fluency tasks (Figure 1).

The highest proportion (approx. 80%) of the pauses was measured in the speech of P2. The result achieved in the task had a little effect on the change of the proportion of pauses in her speech: in spite of her above-average performance in phonemic fluency task, almost the same pause ratio was shown, as in the action fluency task in which she performed quite below average. P1 achieved above-average score in the action fluency task, and his pause ratio was lower than in the other fluency tasks. However, in his speech, there was no significant difference in the proportion of pauses between average semantic fluency and below-average episodic fluency results. In controls, the highest pause ratio was shown in

Table 4. Speech rate and articulation rate in the four speech tasks (sounds/sec).

	Spontaneous narratives	Scenario describing task	Picture describing task	Narrative recalls
		Speech rate		
P1	8.9	8.0	7.0	6.8
P2	8.6	8.0	7.5	6.4
C1	13.2	12.2	12.3	10.6
C2	9.3	9.0	8.3	8.2
		Articulation rate		
P1	12.2	11.6	11.0	9.9
P2	13.0	12.2	12.6	11.6
C1	16.2	15.3	15.0	14.1
C2	12.3	12.3	10.7	11.8

Figure 1. The proportion of pauses in the total speaking time of fluency tasks.

the phonemic fluency task and the lowest in the action fluency task. This result is consistent with the number of produced words: control persons presented the least number of words in the phonemic fluency task and the most in the action fluency task.

Analyzed parameters in the four speech tasks

Table 4 shows the results of temporal measurements and analysis. Different effects of different cognitive resources were manifested in each speaker: spontaneous narratives were characterized by the fastest speech and articulation rates, narrative recalls were characterized by the slowest speech and articulation rates (except articulation rate of C2). The speech rate which refers to speech planning processes more precisely was faster in the speech of the control speakers in every speech task than in the speech of the patients. The difference between

patients and controls is big, since 5% change in speech rate is the just noticeable difference for listeners (Quené, 2007).

In articulation rate mainly referring to the motor component, there was no such difference between speech tasks and between patients and control speakers. The average articulation rate in healthy Hungarian adults is 12.5–14 sounds/sec (Gósy, 2004). This value was produced by P2 and C1 at least in one speech task. In addition, C1 speaks faster than average. Articulation rate of P1 and C2 was below the average Hungarian articulation rate in each speech task. The difficulties of speech tasks were manifested in articulation rate in the two extreme types of tasks, but the difference was not as big as in speech rate. Articulation rates measured in scenario describing task and picture describing task did not show a similar tendency to speech rate in the speech of P2 and C2.

Figure 2 shows the effect of the relative difficulty of speech tasks compared to each other. It shows how speech rate changes in the speech rate of each speaker compared to spontaneous narratives. The difficulty of speech tasks appeared in speech rate. However, the rate of deceleration was higher in each task in the speech of patients than in that of controls.

There were also differences between patients and control speakers and among speech tasks in the pausing strategies (see Table 5). Patients had more pauses, and these pauses were longer in average in each speech task than the pauses of control speakers (see proportion of pauses in the total speaking time, mean duration of pauses, frequency of silent pauses). Speech units of patients were shorter (except spontaneous narratives) than those of controls. However, there was not any difference between patients and controls in the frequency of filled pauses: C2 produced filled pauses the most frequently, while in certain speech tasks, P2 produced them the least frequently.

The difficulty of speech tasks mainly appeared in the frequency of pauses and the proportion of pauses in the total speaking time.

The effect of the complexity of the speech task on the proportion of pauses in the total speaking time is shown in Figure 3. The ratio of pauses is represented in comparison to the spontaneous narratives in each task and in each speaker. Ratios were higher in the speech samples of patients compared to spontaneous narratives, while lower in picture descriptions of control speakers.
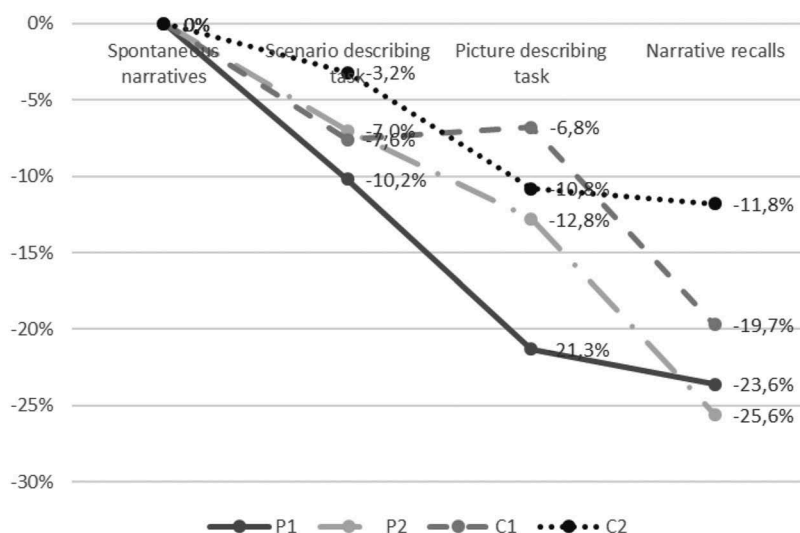

Figure 2. Deceleration of speech rate in each speech task compared to spontaneous narratives.

Table 5. Pausing strategies in the four speech tasks.

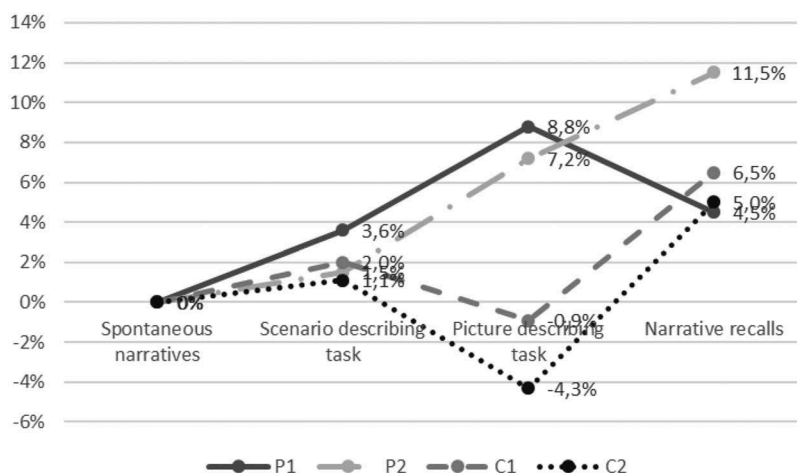|	Spontaneous narratives	Scenario describing task	Picture describing task	Narrative recalls
Proportion of pauses in the total speaking time (%)				
P1	27.2	30.8	36.0	31.7
P2	33.3	34.8	40.5	44.8
C1	18.5	20.5	17.6	24.6
C2	26.0	27.1	21.7	31.0
Mean duration of pauses (ms)				
P1	540	632	632	527
P2	701	784	853	1062
C1	438	533	575	491
C2	455	491	450	490
Mean duration of speech units (ms)				
P1	1667	1576	1441	1383
P2	1728	1471	1214	1414
C1	2222	1703	3052	1902
C2	1621	2592	2379	1546
Frequency of silent pauses (number of silent pauses in 100 words)				
P1	21.3	24.2	27.9	32.3
P2	20.6	20.2	26.2	28.6
C1	13.2	11.0	10.8	17.3
C2	19.1	19.2	17.4	27.8
Frequency of filled pauses (number of filled pauses in 100 words)				
P1	2.9	2.9	5.4	6.5
P2	4.7	2.2	0.9	2.4
C1	2.3	2.6	1.1	5.6
C2	5.7	6.5	7.6	9.7

Figure 3. Changes in the proportion of pauses in the total speaking time.

Discussion and conclusion

In this paper, a two case study was presented from the aspect of the effect of cognitive resources on temporal factors of speech of MS patients. Our hypotheses were partly confirmed.

There were relatively big differences between MS patients and control speakers in the temporal features. Speech rate which refers to speech planning processes more precisely was faster in the speech of control speakers than in the speech of patients. However, articulation rate was similar in the speech of P2 and C2. This shows that the speed of operation of articulation organs is not necessarily different between MS patients and control speakers, while some cognitive functions differ. The proportion of pauses in the total speaking time was lower in controls than in patients. However, it should be noted that all this was true if patients were compared to the age- and gender-matched controls.

The first hypothesis concerned the effect of speech tasks. The hypothesis was confirmed: speech and articulation rate were affected to a different degree by the tasks requiring the use of different cognitive resources. In tasks which require more planning and larger capacity of memory and guided attention, increased cognitive load lead to decreased speech and articulation rate and increased pause duration and frequency. Spontaneous personal narratives which required relatively few cognitive resources were the fastest and in the speech of patients, they contained the lowest proportion of pauses. Speech rate was the slowest in the narrative recall of each participant. This confirms the data in the literature that narrative recall is a complex and dynamic task of cognitive resources that can be a sensitive linguistic tool for the measurements of cognitive processes in the background.

There were major changes in the spontaneous speech of MS patients when performing tasks requiring different cognitive resources than controls (second hypothesis). Comparing the participants by pairs (P1 – C1; P2 – C2), we found that in each task, the proportion of pauses in the total speaking time and the mean duration of pauses in the speech of patients were higher than in that of controls. Also, the slight limitation of speech

planning processes is indicated by the fact that the mean duration of speech units is longer in the speech of controls than in the speech of patients.

According to the third hypothesis, personal narratives were easier for patients than non-personal narratives. The results confirmed our hypothesis. In non-personal speech tasks, a higher rate of speech deceleration was measured in the speech of the patients than in the speech of the control speakers. This confirms with other research findings that the construction of personal narratives and conversational storytelling, which is also mostly personal, requires less cognitive resources (Abraham et al., 2008).

There are specific differences between patients and controls in the picture describing the task. While the speech and articulation rate decelerate in the speech of both the patients and the control speakers compared to the spontaneous narratives, there are different processes between the two groups in pausing strategies. The differences suggest that while visual stimulus facilitates speech planning and execution for the control speakers, the picture description appears to be a more difficult task for patients than the spontaneous narrative.

In both patients, the proportion of pauses in the total speaking time was the lowest in this task. In case of P1, this was the highest, while in case of P2, this was the second highest proportion after the narrative recall. The effect of the visual stimulus is shown by the shortest speech units in the patient's speech. In contrast, the speech planning processes of the control speakers were helped by the visual stimulus, because in this case, we found the longest speech units in this task. We see a parallel pattern in silent pauses, too: control persons produce the least silent pause in this task, while in the speech of patients we found much more silent pauses compared to spontaneous narratives. The patterns shown in this task confirm the observation that visual stimuli in healthy individuals help speech processing and production processes. In addition, the results also confirm the observation that picture description tasks can be a sensitive tool for the recognition of certain neurodegenerative disorders (Choi, 2009; Cuetos et al., 2007). However, exploring the underlying causes of the phenomenon should be the aim of further research.

Comparing the results of the cognitive tests and the performance in speech tasks led us to the following conclusions. Results of case studies show that different test procedures can produce different results in the same cognitive ability. Although the digit span and non-word repetition test theoretically measure the same working memory unit, patient's results were different in the two test: in the digit span test they were slightly (P1) or severely (P2) below the average, while in the non-word repetition test they both achieved average scores according to Hungarian standard. Both patients had a low performance in the Corsi Block Tapping test, which measures the spatio-visual sketchpad. The performance in verbal fluency tasks showed the greatest variability, although they all are applied for measuring the same part of the working memory. Some studies also present a difference between phonemic, semantic, episodic and action fluency (Shao, Janse, Visser, & Meyer, 2014). These results suggest that these tasks require the functioning of partly different cognitive processes. According to Connick, Kolappan, and Bak (2012), the patient's performance in phonemic and category fluency predicts cognitive impairment to a greater or lesser extent. They propose accurate cognitive assessment under 10 words per minute in phonemic fluency and under 20 words per minute in semantic fluency. We found low performance in P2 in the semantic fluency task (11 words, Hungarian normative data in the similar age group: 17). The greatest difference was between the patients in action fluency task: P1 performance was strongly above average (29 words, international

normative data in the similar educational group: 21,), but P2 performance was strongly below average (11 words, international normative data: 21). Several studies suggest that action (verb) fluency requires higher cognitive load than other verbal fluency tasks (Alegret et al., 2018; Woods et al., 2005). Action fluency performance can predict dementalization processes unlike other fluency tasks (Alegret et al., 2018; Mousavi, Mehri, Maroufizadeh, & Koochak, 2014). Although the variability in the verbal fluency tasks was high between the tasks and the patients, the two patients achieved similar results in the other tests of the working memory subsystem.

The Stroop test seems comparable with speech tasks: the increased cognitive load occurs in increased speech production time and decreased the speech rate. This suggests that the Stroop test might be a sensitive tool for the measurements of limitations of cognitive processes in MS.

We can conclude that certain cognitive abilities remained more intact in P1 than in P2, but the attention control tasks had a higher negative effect on the speech of P1 than P2. The results of cognitive tests and the patterns of changes in speech rate and pauses suggest that the cognitive load has a crucial effect on attention, working memory processes and speech production processes. This effect has a greater influence on the speech production of patients with multiple sclerosis than controls.

Limitations

This study has several limitations. The study only uses observations from four cases which did not allow the statistical analysis. Although speech tasks were carefully selected, more tasks are needed to achieve more accurate results. The past records of the neurological and neuropsychological status of the patients were not complete. Neuropsychological anamnesis was not performed during the patients' medical history. The neurological history was well documented in case of P2 but could have been slightly more detailed in case of P1. Appropriate documentation of the patients' medical history would support the interpretation of the results and future research methods and materials.

During interpreting the results, the question arises whether it is sufficient to match the controls with the patients by age and education. The present research shows that it may be worthwhile to match the control speakers to the patients by speech and articulation rate measured in spontaneous narratives because the great differences in the 'ordinary' spontaneous speech rate of the speakers can make the valid interpretation of the results difficult.

Results draw attention to the fact that it is worthwhile to carry out more analysis of the speech of patients with MS. In our further research, we would like to test the results with more patients, testing more cognitive functions.

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Declaration of interest

We hereby declare that we have no financial or personal relationships with other people or organizations that could inappropriately influence the work reported in this paper. We have no affiliation with any organization with a financial interest, direct or indirect, in the subject matter or materials discussed in the manuscript that might affect the conduct or reporting of the work submitted.

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