

Articles

Speech temporal organization in three basal ganglia-related neurological conditions

Organização temporal da fala em três condições neurológicas relacionadas aos núcleos da base

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ABSTRACT

Basal ganglia participate in neural networks that control voluntary body movements, including speech. Diseases that affect the function of these structures can generate abnormal hypokinetic or hyperkinetic movements, influencing speech motor control. How does prosodic temporal organization vary in dysarthria due to Parkinson's disease (PD), Huntington's disease (HD), and Sydenham's chorea (SC)? Three clinical groups (PD with and without medication, HD, and SC) of 15 participants and a control group (n = 18) read a text aloud. Speech fluency measures were related to syntactic boundaries within the text. There was no correlation between global motor scales and temporal parameters of speech. There were correlations between syntactic limits and the duration of pauses in all groups. Only the HD results differed from the other clinical and control groups. Clinical groups are slower to produce speech but preserve the syntactic function of prosody at different levels. Basal nuclei dysfunction appears to affect all clinical groups, regardless of etiology.

Keywords: *dysarthria; speech acoustics; articulation disorders; movement disorders; phonetics.*

RESUMO

Os núcleos da base participam de redes neuronais que controlam movimentos voluntários do corpo, incluindo os da fala. Doenças que afetam a função dessas estruturas podem gerar movimentos anormais hipocinéticos ou hiperkinéticos, influenciando o controle motor da fala. Como a organização temporal prosódica varia na disartria devido à doença de Parkinson (DP), doença de Huntington (DH) e coreia de Sydenham (CS)? Três grupos clínicos (DP com e sem medicação, DH e CS) de 15 participantes e um grupo controle (n = 18) leram um texto em voz alta. As medidas de fluência da fala foram relacionadas aos limites sintáticos dentro do texto. Não houve correlação entre escalas motoras globais e parâmetros temporais de fala. Houve correlações entre os limites sintáticos e a duração das pausas em todos os grupos. Apenas os resultados de DH diferenciaram-se dos demais grupos clínicos e em relação ao grupo controle. Os grupos clínicos foram mais lentos na produção da fala, mas preservam a função sintática da prosódia em diferentes níveis. A disfunção dos núcleos da base parece afetar todos os grupos clínicos, apesar da etiologia.

Palavras-chave: *disartria; acústica da fala; transtornos da articulação; transtornos do movimento; fonética.*

1. Introduction

From an acoustic point of view, we can define *speech* as a nearly continuous sound flow. However, the sound flow is interrupted by numerous silences of variable duration and different origins. Pausing is an essential element in speech; speakers pause to breathe, plan the content of their message, and structure their statement. Also, speech production involves organizing chunks in the speech stream, thus facilitating the parsing of information into meaningful units for the hearer. One of the ways speakers must chunk the speech stream is by placing silent pauses at those places where language units end, that is, at the limits of components in the syntactic structure. Pause time varies from speaker to speaker and by type of speech and represents 40-50% of speaking time for most speakers. Speech speed that is more or less stable in the same individual, pause time, and rate inversely correlate. In this paper, we are interested in the performance characteristics of speech as produced by people with dysarthria, including how people place the pauses in discourse related to syntactic structure.

Darley et al. (1969) defined dysarthria as a group of changes resulting from disturbances in muscle control of the speech mechanism arising from some damage to the central or peripheral nervous system, causing problems in oral communication due to paralysis, weakness, or incoordination of muscles related to speech. Possible changes in dysarthric speech are related to impaired breath support for speech, vocal quality, loudness, breathing pattern, pitch, nasality, consonant and vowel accuracy, phoneme length, pausing, production rate, and emphasis. In dysarthric people, in general, articulation time is increased, as is the time to displace the articulators, with reduced articulatory space (Tjaden & Wilding, 2004).

Alterations in the neuronal circuits involving the basal nuclei can generate abnormal hypo- or hyperkinetic movements, which, consequently, can influence the motor control of speech. How different neurological diseases alter all these parameters is not precisely known. Several studies indicate that any disease that causes alteration in motor control may also cause alteration in the temporal organization of speech. In this paper, we focus on three diseases that cause abnormal

movements: Idiopathic Parkinson's disease, Huntington's disease, and Sydenham's chorea.

Parkinson's disease (PD)

PD is the most common disease with basal ganglia dysfunction. It is characterized by degeneration of neurons in the substantia nigra *pars compacta*, resulting in decreased dopamine in the nigrostriatal fibers. From the functional point of view, two parallel neuronal circuits participate in the control of speech production in PD, one related to the basal ganglia and the other to the cerebellum (Pinto, 2009). In the case of PD, the first influences the second. The dopaminergic deficit characteristic of the disease is responsible for the typical clinical picture: akinesia or bradykinesia (slowness in the initiation and execution of movements, respectively), rigidity, rest tremor, and postural instability (Defebvre, 2005; 2007; Ferraz, 2006; Rosin et al., 2007). Levodopa (L-dopa) is the most effective medication available for PD treatment. However, limitations such as loss of effectiveness, fluctuations (Goberman et al., 2002), motor complications, and mental alterations may arise from long-term use. Dyskinesia (hyperkinesia) is among the motor complications resulting from the use of levodopa. In this case, some patients show that when going from the *off* period to the *on* period, chorea is its main manifestation. That is, these patients go from bradykinetic (when in the *off* period) to hyperkinetic (when in the *on* period) (Defebvre, 2007; Goberman & Coelho, 2002; Hoff et al., 1999; Pinto et al., 2004).

A soft, monotone, breathy or hoarse voice, and imprecise articulation, coupled with decreased facial expressions ("masked face"), contribute to the limitations people with PD have when communicating (Ramig et al., 2008). Changes in speech motor control emerge through inappropriate pauses, hesitations between sequences of movements, an effort to vary melody and accentuation, reduced intensity, difficulty producing speech without interruption, initiating articulation, pausing a continuous response, and occasionally difficulty switching one movement to another. However, these findings may be ambiguous or inconsistent (Angelis, 2006; Martinez-Sánchez, 2010; Mourão & Ferraz, 2003; Pinto et al., 2004; Pinto et al., 2010; Rosen et al., 2006; Sapir et al., 2008; Spencer & Rogers, 2005; Viallet & Teston, 2007). At

the acoustic level, there is a consensus in that the fundamental frequency does not vary as much as in the speech produced by people without PD (Azevedo, 2001; 2007; Duez, 2007; Ramig et al., 2008; Rigaldis et al., 2004; Skodda et al., 2009; Viallet et al., 2003). Sapir (2014) argues that speech changes in Parkinson's patients are due to rigidity and bradykinesia as these features do not seem to fully explain hypokinetic dysarthria in PD. The author suggests alternative explanations for dysarthria in PD, as researchers should include multiple behavioral and physiological factors for the study of speech in this population, such as scaling and maintaining range of motion and effort, pre-programming and initiating movements, sensory and temporal processing, automaticity, emotive vocalization, and attention to action.

Studies comparing perceptual and acoustic changes in speech produced before and after L-dopa intake are frequent in this population, but methodological differences make it difficult to compare all results. Some studies describe improvements in different prosodic aspects (intelligibility, intonation, F0, pitch variation, intensity, intelligibility) after medication (De Letter et al., 2007; Hammen et al., 1994; Meynadier et al., 1999; Pah et al., 2021; Sanabria et al., 2001; Viallet et al., 2002). Others did not identify significant changes in speech variables (duration, intensity, and frequency) during the medication cycle (Cavallieri et al., 2021; Chu et al., 2019; Cushnie-Sparrow et al., 2018; Fabbri et al., 2017; Goberman & Blomgren, 2003; Mignard et al., 2001; Poluha et al., 1998; Rusz et al., 2016; Skodda & Schlegel, 2008; Skodda et al., 2011; Whitfielda et al., 2017).

A systematic review and meta-analysis on the impact of L-dopa on the voice of patients with PD (Pinho et al., 2018) revealed that L-dopa therapy modifies F0 and jitter but does not change vocal intensity in the *on* and *off* phases. Similarly, Brabenec et al. (2017) showed that speech characteristics in PD appear to be mainly related to non-dopaminergic deficits.

Huntington's disease (HD)

HD is an autosomal-dominant type of genetic disorder characterized by an altered huntingtin protein configuration, with trinucleotide (CAG) expansion on the short arm of chromosome 4 (4p16.3), which causes

degeneration of the basal ganglia. The number of CAG repeats ranges from 10 to 29 copies in unaffected individuals, but the HD gene contains 36 to 121 copies (Tasset et al., 2009; Thobois & Peisson, 2007). Neurodegeneration mainly affects the striatum, reducing the activity of the indirect striatal efferent pathway. Chorea is the main manifestation of a movement disorder in HD, but other motor alterations also occur, such as dysarthria, postural instability, gait alterations, muscle tone alterations, dysphagia, and bradykinesia. As the disease progresses, parkinsonism may occur along with muscle rigidity and dystonia (Cardoso, 2009). Cognitive and psychiatric deficits also occur in HD (Cardoso, 2009; Cardoso et al., 2006; Duffy et al., 2007; Ross et al., 2019; Thobois & Peisson, 2007).

Since the choreic movements are unpredictable, hyperkinetic dysarthria is variable and may affect all speech parameters differently. The same patient may produce speech with minimal interference or strongly impaired phonation and articulation (Barkmeier-Kraemer & Clark, 2017; Mourão, 2006; Özsancak, 2007). Rusz et al. (2014a) estimated that 93% of HD patients manifest some degree of speech impairment. Undershooting in consonant and vowel production, long pauses, variable flow of speech, absence of pitch modulation, phonatory impairments, hoarseness, and prosodic changes (monotony, increased or inappropriate pauses, and irregular accentuation) are the main dysarthric changes described in HD (Barkmeier-Kraemer & Clark, 2017; Darley, 1975; Rusz et al., 2014a; Skodda et al., 2014). Other reported speech-related impairments in HD are temporal interruptions of various types in the production of Huntington's patients (Illes, 1989), decreased speech rate (related to the time to process articulation, see Volkman et al., 1992), irregular and slow motor execution (Hertrich & Ackermann, 1994), phonation, oral motor control and prosody (Hartelius et al., 2003; Rusz et al., 2013; Velasco-García et al., 2011). In addition, Murray (2000) demonstrated a high correlation between language tasks, speech, and cognitive abilities in HD patients. Rusz et al. (2014b) and Chan et al. (2019) show that changes in speech occur as early as the prodromal stage.

Diehl et al. (2019) identified four distinct subgroups of speech characteristics in a cohort of HD patients, in which speech rate and dysarthria severity are the variables that most differentiated the groups.

Sydenham's chorea (SC)

SC is the neurological manifestation of rheumatic fever, which, in turn, is a systemic inflammatory autoimmune disease resulting from exposure to antigens of beta-hemolytic streptococcus, a bacteria involved in oropharyngeal infections. The antibodies induced by the streptococcus attack the basal ganglia, more precisely the indirect striatal efferent pathway, causing involuntary movements (Ferraz, 2006). There can be up to four weeks between infection and chorea inception. Typically, the disease has a benign evolution with spontaneous resolution after eight to nine months in most cases. Half of the subjects may evolve with a recurrent or a chronic-persistent form of the disease (Cardoso et al., 1999). Cases in which involuntary movements continue for more than two years despite antichoreic medication are considered persistent. Another frequent sign is a decreased muscle tone, as well as other behavioral, attentional, dysexecutive, and neuropsychiatric manifestations (Beato et al., 2010; Cardoso, 2009; Cardoso et al., 1997; Maia et al., 2005; Tumas et al., 2007). The variability of symptoms suggests selective dysfunction of the frontostriatal circuits (Teixeira Jr. et al., 2005).

In most cases of acute chorea, medication controls motor condition, and it frequently extinguishes speech problems. On the other hand, persistent cases maintain motor alterations longer, even when using medication. However, acute cases are becoming less and less frequent.

A clinical retrospective study reported the incidence of dysarthria in 38% of the evaluated SC patients (Tumas et al., 2007). As in HD, the characteristic dysarthria in SC is hyperkinetic, marked by unexpected variations in pitch and loudness, inadequate pauses, constant or intermittent dysphonia, hyper- or hyponasality, and articulatory imprecision, and slow speech rate due to frequent and long pauses (Barkmeier-Kraemer & Clark, 2017). Angelis et al. (1997) characterized the respiratory, phonatory, resonant, and articulatory speech alterations in fifteen SC patients as predominantly of the upper respiratory type, lack of pneumophonic coordination, vocal murmurs, excessive modulation of pitch and loudness, hypernasality, and articulatory imprecision. Few studies addressed temporal organization and prosody in CS. They describe a tendency to an intensity curve with a descending

pattern, more significant intensity variation, higher initial intensity values, lower final intensity, limited F0 range, and slower speech (Oliveira, 2003; Oliveira et al., 2010).

Only one comparative study investigated whether different aspects of speech are affected in two basal ganglia diseases, PD and HD, and whether there would be different relationships between speech initiation, production, and rate (Ludlow et al., 1987). To this end, they evaluated 12 subjects in each disease group and 12 controls for speech reaction time, syllabic, phrasal and pause duration, and syllable repetition rate. There was no statistically significant difference when comparing the reaction times and syllabic duration of the HD group with the control group or the PD group with the control group. The HD group differed significantly from the control group in sentence duration, pause duration (at an accelerated rate), and syllable repetition rate. The authors argue that different temporal aspects of speech are differentially affected in diseases with impaired basal ganglia, suggesting an independent neurological control for each aspect.

Despite many studies that related neurological diseases to speech disorders, questions remain about the regions of the nervous system that control specific aspects of speech, such as temporal organization and prosody. This study aims to describe how the acoustic parameters of temporal organization of speech vary in subjects with dysarthria, according to different clinical conditions that cause abnormal movements. Specifically, we wanted to know if there were correlations between the motor impairment of each disease (specific scales) and temporal changes in speech. Do any of the speech parameters differentiate the groups? Do choreas of different causes (PD *on* - with dyskinesia, HD and SC) imply similar speech temporal characteristics? Is there a relationship between the frequency and duration of pauses and the syntactic boundaries where they occur?

2. Materials and Methods

The study was approved by the Research Ethics Committee (COEP) of the Universidade Federal de Minas Gerais – UFMG under Certificate n° 258/08. The research was conducted in accordance with

the Helsinki Declaration and informed consent was obtained from all subjects involved in the study.

Participants

Participants in all groups had at least two years of schooling. For the clinical groups, there should be no other clinical condition except the one causing dysarthria, and no record of neurosurgery or visual impairment. Participants did not take any anticholinergic medication with Trihexyphenidyl or Biperiden that may interfere with global cognitive functioning. Participants in all clinical groups underwent motor evaluation and a recording session on the same day.

Clinical groups

Fifteen participants (8 women) with idiopathic PD (according to the UK Parkinson's Disease Society Brain Bank criteria in Hughes et al., 1992) underwent motor examination through UPDRS - Section III (Fahn et al., 1987). The participants were in regular use of dopamine-mimetic medication. They recorded the sound sample twice in the same session: once when they came in after at least 12 hours wash-out period (*off* condition), and then at about one hour after taking medication (*on* condition), when they also underwent dyskinesia assessment (Goetz et al., 1994). For all participants, the examination and recording session took place in the same session to prevent possible side-effects of the wash-out period on daily activities.

Fifteen participants (10 women) with HD had a molecular diagnosis and underwent UHDRS examination (Huntington Study Group, 1996) in the same session before recording. Further analyses excluded participants who scored ≥ 3 in the dysarthria subitem of the UHDRS because their speech was unintelligible.

Fifteen participants (8 women) with Sydenham's Chorea in the persistent form were included. They had to fulfill the modified criteria for an acute rheumatic fever and exclude other causes of chorea (Cardoso et al., 1997; 1999; Teixeira Jr. et al., 2005). Motor examination

followed the UFMG Sydenham's Chorea Rating Scale – USCRS (Teixeira Jr. et al., 2005).

Control group

Eighteen participants (9 women) randomly chosen from the community had no active psychiatric or neurological condition, no previous histories of disorders that still influence cognition, and did not use psychotropic medication.

Recording session

Participants read aloud a text sample in a quiet environment. The noise level ranged from 56 to 64 dB (FAST) and from 53 to 66 dB (SLOW) as measured with an Icel DL-4020 digital decibel meter at three moments of 10 minutes each. The text was the first page (183 words in three paragraphs) of a well-known piece of national child literature, with a structure close to that of the French protocol proposed by Duez (2007). The recordings were made after a silent reading of the text, to minimize possible reading problems and possible disfluencies. A Marantz® PMD 660 digital recorder captured and digitized the audio through a Shure® headset microphone positioned 5 cm from the subject's mouth.

Data analysis

Using the reading-aloud of a text, we wanted to improve the production of silent pauses compared to filled pauses⁶, which should be less frequent. This research strategy helped focus on how people with dysarthria separate syntactic components in the produced speech. The first analysis step consisted of identifying and manually marking the pauses (absence of signal in the spectrogram), associating auditory

6. Although a consistent feature of dysarthric speech, non-silent or secondary pauses (filled pauses, extended syllables/sounds, repetitions, and false starts) typically refer to the cognitive processes involved in spontaneous speech production. We did not consider them here.

and acoustic analysis, regardless of their minimum duration. Every absence of a wave signal (in the visual image), associated with the absence of auditory perception of sound, was considered a pause. We segmented sound wave files into pauses and articulated sequences in Praat (Boersma & Weenink, 2009), according to the criteria by Duez (2005, 2007). When the segment after the pause was a stop, there were two possibilities: if it were a voiced stop, the sound sequence would start at the voiced bar or with a visible explosion; if it were voiceless, it was not possible to separate the occlusion from the preceding pause, or it began with the visible burst if there was one. Each pause was measured in seconds and identified according to the syntactic boundary (Perini, 1996, 2006; Reis et al., 2007):

[P0] a boundary that separates syntagmatic constituents within a longer clause.

[P1] boundary between two phrases

[P2] subordinate or coordinate clause boundary.

[P3] independent clause boundary

[P4] paragraph boundary

[P5] within a phrase

As an example, here is part of the text with the characteristic pause markings:

Numa casinha [5] branca [1], lá [0] no [5] sítio [0] do picapau [5] amarelo [1], mora [0] uma velha [0] de mais [0] de sessenta anos [3]. Chama-se [1] dona Benta [3]. Quem passa [0] pela estrada e [2] a vê na varanda, [1] de cestinha [0] de costura [0] ao colo [1] e óculos [0] de ouro [1] na ponta [0] do nariz [1], segue [1] seu caminho [2] pensando [3]: “que tristeza [2] viver [1] assim tão sozinha [1] neste deserto...” [4]”

We proceeded with the sound file and its respective textgrid in Praat to count each type of pause and sum their durations. Orthographic transcription compares the utterances to the original text to identify disfluencies, repetitions, and omissions and count the number of the phonetic syllables produced. The phonological syllables in the text were identified and quantified, including added, repeated, or omitted. Considering the possible fusion of sounds across word boundaries (external sandhi), 338 is the minimum number of possible syllables.

On the other hand, in the absence of any fusion, the maximum number of syllables would be 362.

The same researcher (THM), who has vast experience in speech-language pathology and linguistics, performed all the analyses. After the analyses, we computed the following measures:

1. Total speech time, comprising total articulation time (sound sequences produced) and total pause time (summed duration of P0 to P5).
2. Number of pauses, and average duration of the pause.
3. Speech rate, calculated by dividing the number of syllables by total speech time.
4. Articulation rate, calculated by dividing the number of syllables by the total articulation time.
5. Total fluency time, which is the duration of speech sequences without disfluency (hesitation, blocking, or repetition).

For statistical analyses, we considered as dependent variables all those related to the temporal organization of speech. The following variables describe the sample characteristics: clinical condition, educational level, age, global motor impairment, and influence of medication (ON and OFF condition). Pearson's correlation coefficient assessed the significance of correlations between variables. For all analyses, the significance level adopted was 5%.

We did not consider the *omnibus* test in the analyses we report, except in the single case of the school years, as we explain below. Instead, we preferred multiple comparisons. The student's t-test with Bonferroni's correction for multiple comparisons assessed for significant differences between the means of each pair of groups. As the sample for each group is not large enough, the Bootstrap method makes it possible to obtain a 95% confidence interval for the parameter evaluated in each test and allows for the estimation of parameters that make up a joint sample originating from the combination of individual samples from each population. We adopted a resampling process of one thousand samples that are replicas of the data with which the test evaluates the variability of quantities of interest with no normality assumption. The temporal parameters may vary with the content

produced and dysarthria's type and severity. As more severe dysarthric patients tend to produce shorter sentences than less compromised patients, the temporal characteristics may be different only because they differ in the length of production and probably also because of a syntactic simplification to achieve the reduction in extension (Bunton et al., 2000). It is possible to build an empirical distribution for the parameters regardless of the data distribution using the Bootstrap method (Efron & Tibshirani, 1994). This empirical distribution is assumed for the parameters and was used instead of the tabulated T distribution. Thus, there is no need for normality and data manipulation, such as removing outliers or transformations.

We hypothesized that the hypokinetic patients had different speech duration patterns than the hyperkinetic ones and that there would be correlations between the motor impairment of each disease (specific scales) and temporal changes in speech.

3. Results

There was no correlation between the different group motor scales and temporal speech organization. Only SC showed a trend towards correlation between the motor scale and articulation rate ($p = 0.08$). Therefore, we do not further mention these results. We next report the variables that characterize the sample, and then the results on the speech temporal organization. Finally, we will arrive at the observed differences in PD's *on*- and *off*-medication conditions.

Sample characteristics

Table 1 shows the obtained values for the variables that characterize the sample. Age is different between the groups as the disease onset is different in each group. This information is not relevant in this study as there are no expected consequences of age on the variables of the temporal organization of speech between about 18-60 years on average as in our samples. As for the school years, we proceeded to the *omnibus* test in this case ($F = 0.302, p = .824$), as it is a desirable result not to have differences that could impact the read-aloud texts we analyzed.

Table 1 – Mean (SD) values for sample characteristics

Variables	Group			
	PD	HD	SC	Control
Age (in years)	56.73 (11.49)	53.40 (14.41)	18.87 (7.39)	27.83 (20.53)
School (in years)	7.9 (4.4)	8.53 (4.91)	7.40 (2.61)	8.44 (2.7)
Years since Diagnosis	8.40 (4.63)	2.96 (1.96)	9.60 (6.97)	-
Motor Rating Scale	<i>Off</i> : 35.8 (10.38) ¹ <i>On</i> : 22.67 (8.08)	40.33 (17.06) ²	5.38 (3.04) ³	-

Notes: ¹ Unified Parkinson's Disease Rating Scale (UPDRS), section III. ² Unified Huntington's Disease Rating Scale (UHDRS). ³ UFMG Sydenham's Chorea Rating Scale – USCRS.

The results show an improvement in the global motor performance in PD after medication. The comparison revealed that the mean difference from the *off*- to the *on*-medication condition (-10.47) is significant (bias = -.038, $p = .009$). In this case, the mean negative value is due to a higher mean value in the *off*- than in the *on*-medication condition.

Temporal organization

We first consider the temporal organization variables: speaking time, articulation time, total pause time, number of pauses, average pause duration, number of syllables, speaking rate, and articulation rate. We then consider the frequency and duration of pauses as related to the syntactic structure. Finally, we comment on the results of the PD group in the *on* versus *off* condition compared to the control group so we may consider a possible effect of medication.

Table 2 – Mean (SD) values for each group and difference between groups

Variables	Group				
	PD <i>off</i>	PD <i>on</i> ¹	HD	SC	Control
Speaking Time (s)	139.25 (87.19)	134.34 (93.48)	204.2 (131.89)	128.6 (43.3)	106.37 (35.13)
Articulation Time (s)	86.14 (26.8)	84.53 (27.44)	107.21 (30.21)	85.94 (19.13)	78.75 (17.07)
Total Pause Time (s)	53.1 (62.52)	49.8 (69.75)	97 (118.66)	42.66 (24.8)	27.61 (19.74)
Number of Pauses	76.93 (58.2)	65.53 (53.1)	99.9 (57.39)	78.4 (36.5)	52.3 (26.87)
Ave. Pause Duration (s)	0.58 (0.21)	0.63 (0.29)	0.82 (0.49)	0.53 (0.11)	0.5 (0.15)
Number of Syllables	370.4 (35.26)	366.26 (37.58)	370.53 (67.55)	377.6 (34.2)	349.72 (11.04)
Speaking Rate (syl/s)	3.25 (1.12)	3.38 (1.13)	2.23 (0.8)	3.19 (0.87)	3.56 (0.92)
Articulation Rate (syl/s)	4.56 (0.97)	4.61 (0.96)	3.6 (0.66)	4.54 (0.76)	4.61 (0.87)

Notes: ¹ Only PD results in the *on*-medication condition were considered for the between-group comparisons.

As Table 2 shows, only the number of syllables is not different between the groups. HD was the most affected group in the temporal organization of speech. HD participants took longer to register the text (204 s) and made more pauses (~100). Their pauses summed up to a longer time (97 s), but each pause was also longer on average (0.82 s). As a result, HD showed slower rates in speaking (2.23 syl/s) or articulation (3.6 syl/s). Post hoc analyses (Table 3) revealed that the differences come mainly from comparing HD and the control group except for speaking and articulation rate, where HD differed from all other groups.

Table 3 – Bonferroni’s multiple comparisons for temporal organization variables between groups

Between-Group Comparison		Variable	Mean Diff.	p-value	Bootstrap ¹			
					Bias	95% Confidence Interval		
					Lower	Upper		
HD	Control	Speaking Time	97.83	.009	-386	40.59	173.5	
		Articulation Time	69.38	.033	-0.485	23.96	137.73	
		Total Pause Time	28.45	.007	0.099	12.62	46.24	
		Number of Pauses	47.6	.02	-0.202	19.27	80.71	
		Ave. Pause Duration	0.32	.016	0.00	0.12	0.62	
HD	SC	Speaking Rate	Control	-1.34	.001	0.004	-1.91	-0.77
			PD on	-0.96	.04	0.015	-1.55	-0.38
			Control	-1.15	.008	0.002	-1.82	-0.51
HD	SC	Articulation Rate	Control	-1.02	.005	0.002	-1.51	-0.53
			PD on	-0.95	.015	0.013	-1.47	-0.43
			Control	-1.01	.008	0.002	-1.62	-0.49

Notes: ¹ Unless otherwise noted, bootstrap results are based on 1,000 bootstrap samples.

Frequency and total duration of pauses at different syntactic boundaries

From Table 4, we see that there were more pauses within a phrase (P5) than at any other boundary, followed by pauses at boundaries between two phrases (P1) and boundaries that separate a subordinate or coordinate clause (P2). The paragraph boundary (P4) elicited the least number of pauses. Differences at the group level appeared at boundaries that separate syntagmatic constituents within a longer clause (P0), at paragraph boundaries (P4), and within a phrase (P5).

Although one may think it syntactically incorrect, a silent pause inserted within a phrase (P5) was the most evident result observed in pauses’ frequency and total duration, both for the control and the clinical groups. Recall that pauses within a phrase are not typical only if we consider the spoken discourse as a linear text, which it is not, even when we have a read-aloud text. We may think of those pauses as critical moments for the readers to plan the content of their message and structure their discourse.

Table 4 – Mean number and average duration (standard deviation) of pauses by type of syntactic boundary

Variable	Boundary Type	Group				
		PD <i>off</i>	PD <i>on</i> ¹	HD	SC	Control
Pause Number	P0	6.67 (6.99)	5.33 (6.11)	10.27 (7.28)	8.2 (4.98)	3.67 (3.46)
	P1	17.67 (9.98)	15.87 (9.08)	20.07 (8.47)	20.07 (8.68)	14.1 (6.26)
	P2	10.93 (2.34)	9.47 (3.58)	11.2 (3.69)	11.8 (3.07)	10.56 (2.2)
	P3	7.13 (1.85)	7.2 (1.15)	6.3 (1.88)	7.6 (1.35)	7.05 (0.94)
	P4	2.13 (0.35)	2.07 (0.29)	1.6 (0.74)	2.13 (0.35)	1.94 (0.24)
	P5	32.2 (41.14)	25.6 (35.25)	51.27 (45.78)	30.13 (23.73)	15.89 (20.96)
Pause Duration (s)	P0	4,14 (5,67)	4,59 (9,19)	9,93 (19,28)	3,42 (2,45)	1,64 (1,82)
	P1	11,44 (11,36)	11,71 (14,11)	18,31 (17,92)	9,83 (4,89)	6,43 (3,93)
	P2	6,46 (4,04)	6,21 (6,49)	9,85 (7,91)	6,82 (3,7)	4,35 (1,36)
	P3	5,76 (3,34)	5,17 (2,77)	6,68 (6,7)	5,05 (1,68)	3,62 (1,0)
	P4	2,97 (1,68)	2,7 (1,56)	2,73 (2,09)	2,05 (0,53)	1,61 (0,46)
	P5	19,74 (40,5)	19,36 (36,7)	49,35 (72,4)	14,36 (15,3)	9,93 (15,9)

Notes: ¹ Only PD results in the *on*-medication condition were considered for the between-group comparisons.

Differences in pause frequency at the group level appeared at boundaries that separate syntagmatic constituents within a longer clause (P0), at paragraph boundaries (P4), and within a phrase (P5). HD participants made more pauses than all other groups at P0 and P5, where they were over three times more frequent than the number of pauses by the control participants. HD pauses were less frequent than all other groups at P4. Differences in average pause duration were at P1, P2, P4, and P5, but the post hoc t-test did not corroborate the difference of total pause duration at P4 (Table 5). Once more, post hoc comparisons show that differences were due to HD performance. HD pause frequency differed from those of controls at P0 and P5, from SC and PD at P4. HD average pause duration differed from the controls at P1, P2, and P5.

Table 5 – Bonferroni’s multiple comparisons for number and duration of pauses by type of boundary (only significant comparisons are shown)

Between-Group Comparison	Variable	Boundary Type	Mean Diff.	p-value	Bootstrap ¹			
					Bias	95% Confidence Interval		
						Lower	Upper	
Control		P0	6.60	.007	.065	2.77	10.71	
HD	SC	Pause	P4	-.53	.008	.015	-.96	-.15
	PD <i>on</i>	Number		-.47	.029	.015	-.87	.08
Control		Total	P5	35.38	.017	-.72	10.99	58.52
			P1	11.88	.028	.01	4.57	22.17
HD	Control	Pause	P2	5.5	.028	.036	2.12	9.86
		Duration	P5	39.42	.048	-.79	7.45	79.60

Note: ¹ Unless otherwise noted, bootstrap results are based on 1000 bootstrap samples.

Finally, results show no difference between the control group and the PD in *on*- or *off*-medication conditions except for the total duration of pauses between paragraphs (P4). Here, a significant difference was detected between the PD group in the *off* condition and the controls (mean difference = 1.37, bias = -.005, $p = .014$). As the results in both conditions were not different within the PD group (a mean difference of only 0.28 s), there was no direct effect of medication. However, the difference to the control group was no longer significant when compared with PD subjects in the *on* condition (mean difference = 1.09, bias = -.009, $p = .065$), an indirect effect of the medication. The PD group was similar in both conditions in all the other speech temporal organization variables and in the frequency and duration of pauses related to the syntactic boundaries where they occur.

4. Discussion

This study investigated how the acoustic parameters of temporal organization vary in the speech of subjects with dysarthria as a function of different clinical conditions causing abnormal movements: Parkinson’s disease, Huntington’s disease, and Sydenham’s chorea. There was no correlation between global motor scales and temporal

speech parameters, which shows that global motor impairment in basal ganglia disorders does not involve speech. Except for the number of syllables, all temporal measures are different when comparing the clinical and control groups. Nevertheless, these differences come mainly from comparing HD and the control group. HD was the most affected group in the temporal organization of speech.

The temporal change may vary with the content produced, the type and intensity of the dysarthria. Since more severely dysarthric patients tend to produce shorter sentences than less compromised patients, the temporal characteristics may be different just by differing in the length of production, and probably also by syntactic simplification to achieve the reduction in length (Bunton et al., 2000).

Parkinson's disease

Longer pauses may be related to two typical alterations of this disease: akinesia or bradykinesia (slowness in the initiation and execution of movements, respectively) and rigidity (Defebvre, 2005, 2007; Ferraz, 2006; Rosin et al., 2007). Disregarding gender and medication, the speech and articulation rates (\pm standard deviations) of the sample studied here are equivalent to those of Duez (2005) and Reis et al. (2007), i.e., slightly lower than those of the control group. These results corroborate previous studies (Azevedo et al., 2003; Goberman and Elmer, 2005; Teixeira, 2008; Volkman et al., 1992), but others observed that speech rate is faster in PD than in control participants (Hammen & Yorkston, 1996; McRae et al., 2002). As Skodda et al. (2010) showed, patients with PD tend to accelerate the pace during movement execution; we assume that speech and articulation rate values tend to obscure this behavior because longer and shorter durations are added together in the mean values.

Not all published studies compare *on* versus *off* conditions or between males and females, but in all of them, participants with PD had a longer average duration of pauses than those in the control group (Duez, 2005; Lowitt et al., 2018; Reis et al., 2007). Lowitt et al. (2018) observed that predictors of group performance in the reading task were percent of utterance duration composed of vocalic intervals

and rate. The patients tended to speak at a slower rate than the healthy controls. Even mildly affected PD differed from controls in their rhythmic performance. The authors observed that spontaneous speech is potentially more sensitive to speech problems in speakers with mild hypokinetic dysarthria than a read-aloud text. Reis et al. (2007) found that the mean duration of pauses in controls was significantly shorter than PD in the off and on conditions. In our study, the average duration of pauses here did not significantly differ between the control and the PD group off and on conditions.

The difference between the pauses within each group shows the behavior in relation to syntactic structure. The only difference between the PD group in the *off* condition and the controls was in the total duration of pauses. This result suggests that the syntactic function of prosody is not altered in PD, as also stated by Duez (2007). We agree with Duez (2005) when she points out that there is more considerable variability in the temporal aspects of speech among subjects with PD.

Studies comparing perceptual and acoustic changes in speech produced before and after L-dopa intake are frequent in this population. However, methodological differences make it difficult to compare all results. Our results identified no significant change in speech measures during the medication cycle. It is known that there is a close correlation between dopamine levels in the striatum and UPDRS values. Therefore, a possible explanation for the lack of improvement in speech parameters after taking the medication is that dopamine is unimportant for speech control. Furthermore, we believe that non-linguistic tasks may better show this speech-specific motor performance, as difficulty in language processing may also influence these results.

As mentioned above, we found no correlation between motor performance and speech parameters in any group. Although they did not use correlation analysis, two studies about medication effect on speech parameters showed similar results to those found here (Skodda & Schlegel, 2008; Viallet et al., 2002). However, they observed no significant improvement in temporal measures of speech after medication, despite the evident global motor improvement as measured by the UPDRS. This result can be interpreted as a lack of correlation between motor scales and speech parameters, confirming Teston and

Viallet's (2005) opinion that using only the single speech item of the UPDRS is insufficient for an in-depth description of speech.

Huntington's disease

Our findings on speech timing agree with previous studies reporting the occurrence of variable and slow rate, prolonged intervals, and inappropriate silences in the speech of HD subjects (Hartelius et al., 2003; Hertrich & Ackermann, 1994; Illes, 1989; Rusz et al., 2014a; Skodda et al., 2014; Vogel et al., 2012). As others before (Illes, 1989; Hertrich & Ackermann, 1994; Skodda et al. 2014), we observed that motor execution is irregular and slow in dysarthric individuals with HD. Our results show that total speaking time, total pause time, total articulation time, number of pauses, average pause duration, and number of syllables are much longer in subjects with HD than in controls. However, speech and articulation rates are lower in these participants.

Unlike our results, Ludlow et al. (1987) showed that the mean duration of pauses in subjects with HD at different stages of the disease was not statistically different from controls. However, these authors measured pauses in a single sentence, and such a small sample most likely influenced their results. Moreover, Volkmann et al. (1992) observed a decrease in speech rate in HD without an increase in pause duration, regardless of disease progression.

Slower articulation rate, imprecise vowel articulation and excess intensity variations were also the most salient patterns of speech dysfunction in the patients with HD studied by Rusz et al. (2014a). However, the authors also found a decrease in the number of pauses.

Murray (2000) observed that the proportion of simple sentences produced by subjects with HD was negatively related to their motor speech skills. Hartelius et al. (2003) described the speech impairments in HD as being in phonation, oral motor control, and prosody, and these were directly proportional to disease impairment. Skodda et al. (2014) showed that speech rate and pause ratio showed correlations to overall motor impairment. Our data revealed no correlation between global motor impairment and speech.

Chorea and other motor deficits may occur in this population, such as changes in muscle tone and bradykinesia (Cardoso, 2009; Cardoso et al., 2006; Duff et al., 2007; Thobois & Peisson, 2007). With the progression of the disease, they may present parkinsonism, muscle rigidity and dystonia. Thus, we expect significant changes in speech production, as revealed by the results found here. Wherever we found a difference in our results, it was due to HD participants. Regarding syntactic boundaries, however, the results show that only part of the syntactic function of prosody is adequate.

Sydenham's chorea

As in HD, dysarthria in SC is hyperkinetic and marked by unexpected variations in pitch and loudness, inadequate pauses, constant or intermittent dysphonia, hyper- or hyponasality, articulatory imprecision, and slow speech rate due to frequent and long pauses (Barkmeier-Kraemer & Clark, 2017). Our results show that total speaking time, total pause time, total articulation time, number of pauses, and number of syllables are much longer in subjects with SC than in controls. The average pause duration is also greater, but only slightly so. Speech and articulation rates are slightly lower in those with SC. Our findings are consistent with previous studies (Oliveira, 2003; Oliveira et al., 2010) which also report slower speech in this population, with speech parameters closest to those of the control group.

We believe that a possible hypothesis for the decrease in speech and articulation rates in SC patients may be the presence of bradykinesia induced by antidopaminergic drugs, commonly used in this population, or a nigrostriatal dysfunction, as already demonstrated (Oliveira et al., 2010; Teixeira et al., 2003). Because HD is different from the other groups in most temporal parameters, one can hypothesize that this is due to neurophysiological issues. However, the SC should have the same response pattern since it is also a hyperkinetic disease. Another option would be the apparent more significant variability of motor impairment in the HD group. However, our analyses did not show a significant change in this respect. Therefore, this difference may be related to the more significant variance in the HD group.

Ludlow et al. (1987) discuss that the fact that different temporal aspects of speech are distinctly affected in diseases that impair the basal ganglia may suggest an independent neurological control for each of these aspects. However, it is also considered that, despite having different neurophysiological causes, the core of the different diseases considered here resemble each other in many temporal aspects regarding motor speech behavior. It is agreed with Oliveira et al. (2010) that alterations in the basal ganglia generate a motor pattern of speech control that is similar, regardless of the cause of the dysfunction.

Regarding syntactic boundaries, the results show that only part of the syntactic function of prosody is adequate.

5. Conclusions

Participants in the clinical groups are slower in speech production but preserved part of the syntactic function of prosody at different levels. It seems that basal ganglia dysfunction affects temporal organization of speech in all clinical groups despite etiology. Compared to PD and SC, HD was the most affected group; when there was a difference between the groups, it was due to HD's results.

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Conflict of interests (multiple authors)

The authors declare they have no conflict of interest.

Credit Author Statement

We, Thais Helena Machado, Ana Cláudia Pereira Bertolino, Leandro Pereira, Francisco E. C. Cardoso, Rui Rothe-Neves, hereby declare that we do not have any potential conflict of interest in this study. Thais Helena Machado, Francisco E. C. Cardoso, and Rui Rothe-Neves have participated in study conceptualization, methodology, study design and data validation and editing. Thais Helena Machado have done data collection and data generation. Thais Helena Machado and Ana Cláudia Pereira Bertolino have done formal data analysis. Rui Rothe-Neves and Leandro Pereira have performed statistical data analysis. Francisco E. C. Cardoso and Rui Rothe-Neves have participated in project administration and project supervision. All authors approve the final version of the manuscript and are responsible for all aspects, including the guarantee of its veracity and integrity.

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