PERSISTENT DEVELOPMENTAL STUTTERING AS A CORTICAL-SUBCORTICAL DYSFUNCTION

Evidence from muscle activation

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Abstract – **Background:** One contemporary view of stuttering posits that speech disfluencies arise from anomalous speech motor control. **Purpose:** To verify the rest muscle tension and speech reaction time of fluent and stuttering adults. **Method:** 22 adults, divided in two groups: G1 – 11 fluent individuals; G2 – 11 stutterers. Electromyography recordings (inferior orbicularis oris) were collected in two different situations: during rest and in a reaction time activity. **Results:** The groups were significantly different considering rest muscle tension (G2 higher recordings) and did not differ when considering speech reaction time and muscle activity during speech. There was a strong positive correlation between speech reaction time and speech muscle activity for G2 – the longer the speech reaction time, the higher the muscle activity during speech. **Conclusion:** In addition to perceptible episodes of speech disfluency, stutterers exhibit anomalies in speech motor output during fluent speech. Correlations with a possible cortical-subcortical disorder are discussed.

KEY WORDS: speech, stuttering, electromyography, reaction time.

Gagueira persistente do desenvolvimento como disfunção córtico-subcortical: evidências pela ativação muscular

Resumo – **Introdução:** Atualmente considera-se que as disfluências da fala na gagueira sejam decorrentes de controle motor anormal. **Objetivo:** Verificar o repouso e tempo de reação para fala em adultos fluentes e gagos. **Método:** 22 adultos, divididos em dois grupos: G1 – 11 fluentes; G2 – 11 gagos. Os dados eletromiográficos (orbicular dos lábios inferior) foram obtidos em duas situações: repouso e atividade de tempo de reação. **Resultados:** Os grupos apresentaram diferenças significantes para a tensão muscular de repouso (G2 valores maiores) e não se diferenciaram quanto ao tempo de reação e atividade muscular de fala. Houve correlação positiva entre o tempo de reação e a atividade muscular de fala para G2 – quanto maior o tempo de reação maior a atividade muscular de fala. **Conclusão:** Além dos episódios perceptíveis de disfluência, gagos apresentam alterações no *output* motor de fala durante a produção da fala fluente. Correlações com possível distúrbio cortico-subcortical são discutidas.

KEY WORDS: fala, gagueira, eletromiografia, tempo de reação.

Speech motor control is related to the systems and strategies that control speech production. In general terms, the input of this system is the phonological representation of language, composed mainly by sequences of abstract units like the phonemes. This input is transformed into articulatory movements that convey the linguistic message through an acoustic signal, which then can be interpreted by the listner[1]. Speech can be defined as a task that requires rapid motor control. The movements involved in speech production are processed, according to data obtained through acoustic analyses, in milliseconds[2,3]. Involuntary speech disruptions occur in persistent developmental stuttering (PDS) and are characterized mainly by sound and syllable repetitions. Stuttering occurs especially during the production of spontaneous speech. Singing, speaking in chorus and in a predetermined rhythm (to a metronome), that is, speech that has its rhythm controlled by an external stimulus is better produced than sponta-

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neous speech. The stutterer speaks more easily when the content of his speech is automatic or hyper-learned[4]. A few studies have focused on the physiology of stuttering, especially in adults, with the purpose of investigating the predisposition, the etiologic factor or even describing not only fluent, but also disfluent speech. A variety of physiological measurements (especially through electromyography, surface and needle) have been used during the moments of stuttering and during the production of fluent speech in stuttering individuals[5,6]. These measurements, however, are in great part influenced by the increase or decrease in the levels of muscle activity, being this type of motor reaction usually associated to the speech of stuttering adults. Besides that, high levels of muscle activity are frequently observed prior to speech onset[7]. This has a negative impact on the highly precise adjustments necessary for the production of speech, which in turn influences the speech performance of stuttering individuals[8].

The onset of speech has been studied in tasks involving the measurement of speech reaction time (RT)[9,10]. Although a great variability is observed between subjects, this has been described as increased in stutters depending on the type and complexity of the verbal task[11]. Several authors have suggested that PDS can result from difficulties in the timing of speech movements, resulting in a poor maintenance and sequencing of speech[12,13]. In the case of stuttering, one of the major issues related to the RT refers to the extension and complexity of utterances. Longer utterances are more complex in terms of motor planning. This increases the difference in the RT of individuals with stuttering[11] once these individuals present difficulties in speech motor planning. Kleinow and Smith[14], point that the extension of an utterance affects the speech motor planning of stuttering individuals, as well as their motor performance. Therefore, changes in the motor performance as a reaction to the increase in the extension of utterances can contribute to the increase in the variability of movement execution as an attempt to reach an adequate production. The influence of the type of task is also observed. Based on the study of Peters et al.[9] and on the considerations of Klapp[10], the RT is greatly influenced by the type of stimulus given at the beginning of processing. Activities involving repetition and reading allow the pre-programming of most motor commands prior to the act of speaking (simple RT). This reduces the effect of complexity and consequently decreases the number of disruptions. During spontaneous speech pre-programming is not possible, at least not for the whole utterance – speech begins before the linguistic planning is completely ready[10]. In this case, one can assume that longer RTs originate from an absence of pre-programming and, therefore, have a greater chance of presenting failures. Overall, studies that investigate stuttering behavior through speech reaction time have found differences, for the same task, between stuttering and fluent individuals. However, these differences are not always related to longer RTs, but to the variability between the data (dispersion of data) presented by stuttering individuals[13]. This variability can indicate that the speech system of stuttering individuals requires several attempts in order to reach a fluent pattern.

The aim of the present study was to verify the rest muscle tension and speech reaction time in fluent and stuttering individuals. The research hypotheses were:

H1 – rest muscle tension is higher in stuttering than in fluent individuals;

H2 – the reaction time for speech onset is higher in stuttering than in fluent individuals;

H3 – muscle activity involved in speech is higher in stuttering than in fluent individuals;

H4 – a positive correlation exists between the reaction time for speech onset and the muscle activity involved in speech production for both groups – the longer the reaction time, the higher the muscle activity.

METHOD

Participants

This study received prior approval of the Institution's Ethics Committee (CAPPesq – HCFMUSP 1021/03) and informed consent was obtained from all of the participants.

Participants of this study were 22 adults, divided in two groups: GI had 11 fluent individuals, 4 female and 7 male, with a mean age of 31:5 years and with no history of communication disorders, hearing loss and neurological and/or cognitive deficits; GII had 11 stuttering individuals, 4 female and 7 male, with a mean age of 25:1 years, with no other associated communication disorder, hearing loss and neurological and /or cognitive deficit, and with no history of previous speech and language therapy.

The adopted inclusion criteria for both groups involved the application of the Stuttering Severity Instrument[15] (SSI-3):

a) GI – to present up to 10 points on the SSI-3, that is, to present no signs of stuttering;

b) GII – to present 11 points or more on the SSI-3, that is, to present at least a very mild level of stuttering.

Procedure

Electromyography – Biological signals were obtained using a four channel module (EMG System do Brasil LTDA), consisting of a signal conditioner with a band pass filter with cut-off frequencies at 20–500 Hz, an amplifier gain of ×1000 and a common mode rejection ratio >120 dB. All data were processed using specific software for acquisition and analysis (AqData), a converting plate for A/D 12 bits signal to convert analog to digital signals with sampling frequency of anti-aliasing 2.0 Hz for each channel and an input average of 5 mV. Differential superficial disposable electrodes (Medtrace Mini Ag/AgCl – 10 mm diameter), separated by 1 cm, were used to capture muscle activity.

Table 1. Descriptive analysis of muscle rest tension (uV).

	Mean	SD	Median	Variation coefficient	p value
GI	16.46	3.66	16.81	22.21	0.02*
GII	26.65	10.98	28.25	41.19	

GI, fluent individuals; GII, stuttering individuals; SD, standard deviation.

Table 2. Descriptive analysis of speech reaction time (milliseconds).

	Mean	SD	Median	Variation coefficient	p value
GI	82.18	8.52	83.00	10.37	0.32
GII	97.7	40.7	99.00	41.69	

GI, fluent individuals; GII, stuttering individuals; SD, standard deviation.

Table 3. Descriptive analysis of speech muscle activity (uV).

	Mean	SD	Median	Variation coefficient	p value
GI	68.45	25.10	64.24	36.67	0.32
GII	75.90	32.35	71.30	42.62	

GI, fluent individuals; GII, stuttering individuals; SD, standard deviation.

Table 4. Correlation between speech reaction time and speech muscle activity.

	r	p value
GI	0.218	0.60
GII	0.716	0.03*

GI, fluent individuals; GII, stuttering individuals; SD, standard deviation.

The EMG surface electrodes are pre-amplified with factor 20 on the electrode itself and factor 50 on the amplifier.

In all procedures the capture and analysis of EMG signals were carried out as recommended by the International Society Electrophysiology Kinesiology (ISEK).

Only one recording channel of the electromyography equipment was used. Muscle activity was captured by disposable electrodes fixed on the middle portion of the inferior perioral region (inferior orbicularis oris), 2 mm below the free margin of the lip[16]. A pair of electrodes was fixed to the skin using adhesive tape (transpore 3M) with a distance of approximately 10 mm between each electrode. Prior to positioning the electrodes, the skin was cleansed with alcohol (96º GL).

Prior to data gathering, participants were asked to sit comfortably on a chair with their heads oriented according to the horizontal plane of Frankfort, parallel to the ground, with their arms resting on their thighs. The reference electrode was placed on the right wrist of the participants.

Testing situations consisted of: a) muscle rest tension – each participant was instructed to remain the more relaxed as possible for 1 minute. After that, five seconds of muscle activity was recorded; b) speech reaction time – each participant was instructed to repeat the phrase *"Barco na água"*[3] (boat on water) as soon as they heard a high pitched beep – indicating the start of the chronometer start. Only fluent productions, free of disruptions, were accepted. The recording of the muscle activity coincided with the start of the chronometer. The speech of each participant was recorded by equipment in five seconds windows.

Analysis of the results

A total of 44 electromyographic recordings were analyzed. The gathered data were quantified in mean root square (RMS) by the signal gathering and processing program and expressed in microvolts (uV).

For the rest condition, the obtained values represent the mean (RMS) electromyographic activity obtained in five seconds. Speech reaction time was obtained using a digital chronometer that measured the time interval between the instruction given to start speech and the actual production of speech. Muscle activation during each repetition of the target phrase was also analyzed. An interval marker was used in order to select the information intervals which were representative of the beginning and end of the muscle contraction (on and off situation) and therefore corresponded to the repetitions of the target phrase. The obtained values represent the average (RMS) electromyographic activity observed during the utterance of the target phrase.

In order to compare the results between participants, EMG amplitude values for each participant were normalized to the highest recorded values (% of max value for each electrode arrangement).

Since the analyzed variables do not present a normal distribution, the Mann-Whitney non-parametric test ($p \leq 0.05$) was used to compare the performance of both groups. The Pearson Correlation test ($p \leq 0.05$) was used to correlate data of speech

reaction time and speech muscle activity. For this analysis discrepant values were discarded.

RESULTS

The obtained results indicate that the groups differed significantly in terms of the rest muscle tension – GII presented higher values (p=0.02). Besides that, for GII there was a strong positive correlation between speech reaction time and speech muscle activity. For this group, the longer the speech reaction time, the higher the muscle activity involved in speech production (p=0.03).

There was no statistically significant difference between the groups for speech reaction time (p=0.32) and for speech muscle activity (p=0.32).

Tables 1, 2 and 3 present the descriptive analyses of the rest muscle tension, speech reaction time and speech muscle activity. Table 4 presents the correlation between speech reaction time and speech muscle activity.

DISCUSSION

In the present study, high levels of muscle activity during the rest condition, verified in individuals with PDS, agrees with the findings of McClean[6]. This result suggests that high levels of input in the mechanoreceptors pathways during inadequate moments (e.g. prior to speech) can have the effect of disrupting neuromotor activity patterns and suggest muscle hyperactivity probably of a suprasegmentar origin. Also, the data found for the stuttering individuals were uniform. For this group, the results regarding rest muscle tension were twice as disperse than that obtained for fluent individuals. This confirms the first hypothesis of this study.

The comparison with other nosologic entities related to speech motor control disorders, especially spasmodic dysphonia (SD), a type of laryngeal dystonia, is pertinent. In SD physical concomitants are common during speech due to the effort made to speak and possibly due to an associated orofacial dystonia[17,18]. In dystonias secondary to subcortical brain injuries, muscle co-contractions frequently occur, i.e. agonists and antagonists are simultaneously activated with the expansion of the contraction to adjacent muscles[2,19,20]. In SD, interruptions in voice production occur mainly during speech and less frequently during simple vocalizations and singing[2,20]. Positive family history is significantly higher in individuals with torsion dystonia than in the general population[21]. Specific task dystonia or "occupational cramp" is a focal dystonia that compromises rapid and automatic motor sequences. Stuttering can be defined as a highly specific disorder, limited to speech.

Electrophysiologic studies of dystonia have demonstrated a hyper excitability of medullar motoneurons and of the brainstem[22]. These abnormalities are present in regions that are not clinically affected by the disorder (focal dystonia). The study of finger movements in stutteres suggests a significant statistical difference between stutterers and fluent individuals. This difference is not restricted to the laryngeal and orofacial muscles, but compromises the motor systems that are not related to speech[23].

Regarding speech RT, the data of this study agrees with the findings of Bosshardt et al.[13] who found no differences between suttering and fluent individuals and with the data of Kleinow and Smith[14] who found a greater variability in the temporal patterns of stuttering individuals (in the present study the group of stutterers presented results four times more disperse than their controls). This variability, indicated by the dispersion of the data, suggests that the speech system of stuttering individuals requires several attempts prior to speech in order to reach a fluent pattern. Given this, the second hypothesis of the study was not confirmed.

Speech muscle activity involved in the speech RT task, although not expected, did not differ between suttering and fluent individuals. The similar myoelectric activity observed for both of the studied groups indicates the effort made by stuttering individuals to maintain fluent speech, i.e. for these individuals a greater motor control was necessary to produce a speech free of disruptions – in this task only fluent productions were accepted[3].

During data gathering, for the group of stutterers, several repetitions of the target phrase were necessary in order to obtain a speech motor pattern free of disruptions. The changes required to maintain speech fluency, associated to the level of compromise of the motor processes involved in speech production, can be related to the several repetitions necessary to adapt the speech system and avoid disruptions. Possibly, the sutterers of this study present difficulties to initiate and program speech which was facilitated by motor learning. Given these considerations, the third hypothesis of the study was not confirmed.

Regarding the fourth hypothesis of the study – partially confirmed – the positive correlation verified between speech RT and speech muscle activity for the stuttering group suggests the impact of poor timing over the motor system. Several authors point that high muscle activity makes speech more vulnerable to disruptions[1,3,6,14,15].

The initiative of a movement, including speech, depends on brain structures associated to movement. The cingulum gyrus and the supplementary motor area are important to motivated behavior. The anterior cingulum circuit receives information of other limbic structures, projects itself to the ventral striatum, entering the cortico-subcortical loop along with other basal ganglia[24-26].

The pre-motor area is related to movement planning, along with the support of the cortico-subcortical loop[24]. Movements that initiate as a consequence of internal motivation involve the supplementary motor area. Movements that arouse from external events involve the lateral pre-motor area.

The mesial pre-motor area is involved in the execution of motor sequences. Studies developed with monkeys indicate that neurons of the globus pallidus deflagrate before the end of the sub movement in an automatic and predictable sequence[27]. It has been proposed that this activity is an internal cue that indicates the final motor component in a sequence of movements. This cue would be sent to the supplementary motor area responsible for executing the change to the next movement in an organized sequence[21,27]. According to this model, the first segment of a motor sequence would begin in the motor cortex and the basal ganglia would control the subsequent motor segments.

The supplementary motor area is especially involved in self-initiated and sequenced movements. Its function is mainly related to the temporal aspects of movements. As already discussed, speech requires a rapid and sequential motor control and is processed in milliseconds, requiring a high temporal resolution. The supplementary motor area creates internal time cues to facilitate the beginning of sub movements in a well learnt motor sequence[21,28].

Motor learning involves the repetition of the movement and the activation of the pre-motor area. It has also been suggested that the cerebellum is critic for motor learning[29]. Learning increases the automatic component of a voluntary motor act. In oral verbal productions, automatic sequences also exist. Combinations of groups of muscles related to more frequent articulatory movements in a certain language also become automatic.

Stuttering individuals present a greater compromise of spontaneous speech, which has a particular rhythm. RT increases, suggesting a difficulty with the onset of motor programming. During the maintenance and sequencing of speech, there is a difficulty to skip from one motor pattern to the next. In stuttering, repetitions occur mainly on the first sound or syllable of a word, indicating that the dysfunction of the basal ganglia does not indicate the end of the first component which is repeated, breaking the sequence[21].

This model presents a possible mechanism for the effect of rhythm in stuttering: the external cue compensates the deficit of the internal cue. When reading in chorus and singing, the voice of the other person propitiates an external temporal cue[21].

Neurogenic stuttering has been described in injuries of almost all brain regions, except of the occipital lobe[21,30]. However, in 75% of the cases documented by neuroimaging, the injury is subcortical[2], of the basal ganglia, internal capsule and corpus callosum, and of the frontosubcortical loop. Thalamic stimulation[21,31] and of the supplementary[21,24] motor area of vigil surgical patients produce the repetition of the first syllable of words.

Studies with functional magnetic resonance imaging and positron emission tomography in stutterers indicate a higher activation of anterior areas, in the primary motor cortex, supplementary motor area, anterior cingulum and cerebellum, that is, areas related to the initiative, planning and sequencing of movements[12,30]. The activation of temporal language areas is totally absent. A greater participation of the right hemisphere is also observed[12].

Magnetoencephalography reveals that the sequence of brain activation differs in stuttering and fluent individuals. In fluent speakers, the activation of the left inferior frontal region precedes the activation of the motor and dorsal pre-motor cortexes. In sutterers, the sequence is reversed, with abnormally early responses in the left motor and pre-motor regions, followed by the activation or the left inferior frontal cortex. This indicates that the stutterer seems to initiate the motor program before the articulatory programming[8].

Finally, it is possible to state for PDS the existence of high levels of muscle activity due to a suprasegmentar influence, and a difficulty with motor initiative, motor planning and sequencing of speech movements. The basal ganglia influence muscle tonus, the preparation and selection of a motor plan. Similarities between PDS and focal dystonias are attractive. Stuttering can be seen as resulting from a cortico-subcortical disorder, where there is no alteration of a particular brain region, but a dysfunction of a system that interferes with the rapid and dynamic processing of speech.

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