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Indicators of speech fluency in stuttering and in phonological disorder

Indicadores de fluência da fala na gagueira e no transtorno fonológico

Keywords

Stuttering
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Phonological Disorder
Speech
Comorbidity

Descritores

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ABSTRACT

Purpose: To identify the indicators of speech fluency that differs subjects with stuttering, with phonological disorder, and with the two disorders in comorbidity. **Methods:** Thirty subjects aged 4-11 years old were divided into 3 groups, each one with 10 subjects: groups with developmental stuttering (SG), phonological disorder (PDG), and with two diagnoses in comorbidity (SPDG) participated in this study. The procedures were speech fluency and phonology evaluation. The data were submitted to statistical analysis. **Results:** Subjects from SG and SPDG showed a greater occurrence of stuttering-like disfluencies and total of disfluencies in relation to the subjects with PDG. Regarding to the other disfluencies, the three groups were similar. Subjects with PDG showed fewer monosyllabic word repetitions, part of word repetition and prolongations in relation to subjects from SG and SPDG. Blocks occurred more frequently in the two groups with stuttering (SG and SPDG) than in the group with PDG. Interjection occurred more frequently in subjects from SG than in PDG. **Conclusion:** The PDG was the most differentiated in quantitative and qualitative terms in the three groups analyzed. The similarities and differences between the groups will assist the differential diagnosis and, consequently, will enable improved therapy. The presence of blocks represents an important marker for the diagnosis of stuttering

RESUMO

Objetivo: Identificar os indicadores de fluência da fala que diferenciam os sujeitos com gagueira, com transtorno fonológico e com os dois distúrbios em comorbidade. **Método:** Participaram deste estudo 30 sujeitos de 4 a 11 anos, separados em 3 grupos, cada um com 10 sujeitos: grupo com gagueira do desenvolvimento (GG), transtorno fonológico (GTF) e os dois diagnósticos em comorbidade (GGTF). Os procedimentos foram: avaliação da fluência da fala e da fonologia. Os dados foram submetidos à análise estatística. **Resultados:** Os sujeitos do GG e GGTF apresentaram maior ocorrência das disfluências típicas da gagueira e do total das disfluências em relação aos do GTF. Em relação às outras disfluências, os três grupos foram semelhantes. O GTF manifestou menor quantidade de repetições de palavra monossilábica, de parte de palavra e prolongamentos em relação aos sujeitos dos GG e GGTF. Os bloqueios ocorreram mais frequentemente nos dois grupos com gagueira (GG e GGTF) em relação ao GTF. A interjeição ocorreu com maior frequência no GG quando comparado com o GTF. **Conclusão:** Dos três grupos analisados, o GTF foi o que mais se diferenciou em termos quantitativo e qualitativo. As semelhanças e diferenças entre os grupos auxiliarão o diagnóstico diferencial e, consequentemente, possibilitarão melhor terapia. A presença de bloqueio representa um importante marcador para o diagnóstico de gagueira.

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INTRODUCTION

Stuttering is a childhood-onset neurodevelopmental, complex and multifactorial fluency disorder, characterized by frequent disruptions in the flow of speech⁽¹⁾. Linguistic, cognitive and motor skills may be related to the disorder⁽²⁾, and especially the phonological variable has received considerable attention in the literature⁽³⁾.

The Covert Repair Hypothesis theory⁽⁴⁾ proposes that delayed phonological encoding in persons who stutter leads to an increased internal correction of coding errors, causing stuttering events. It is also believed that higher phonological demand impairs the stability of the vulnerable motor system in persons who stutter⁽²⁾. Therefore, some stuttering theories suggest that difficulty in phonological encoding may delay or disrupt planning and subsequent articulatory execution, causing disfluencies⁽⁵⁾.

Briefly, phonological disorder in children who stutter co-occurs at a substantial rate: it is estimated that from 16 to 30% of children who stutter also have phonological disorder, while the prevalence in the general population is from 6 to 8%^(6,7). Despite the frequent reference in literature to the coexistence of disfluencies in children with phonological disorder, there are few studies that describe in detail the manifestations of disfluencies that may differentiate these two conditions⁽⁷⁾.

In order to highlight the co-occurrence of phonological disorder and stuttering, a study showed that children with phonological disorder often have disfluencies or hesitation marks, for example hesitations, reformulations, repetitions, prologations, silent and filled pauses, beyond false beginnings⁽⁸⁾. However, although the authors of this research describe the presence of disfluencies in children with phonological disorders, the interpretation given to disfluencies is that they would be constitutive in the process of phonic acquisition⁽⁸⁾.

Although studies indicate the presence of disfluencies in children with phonological disorders^(7,8), the number of investigations that deepen the analysis of disfluencies is restricted, and consequently little is known about similarities and differences between the disruptions that occur in these two disorders.

In particular, it is assumed that: (H1) subjects with stuttering and subjects with both disorders in comorbidity would have a higher frequency of occurrence as regarding the stuttering-like disfluencies as in relation to other disfluencies compared to those with phonological disorder; (H2) Regarding the typology of disfluencies, there would be differences between subjects with phonological disorder compared to subjects with stuttering and those with stuttering and phonological disorder in comorbidity, that is, subjects with phonological disorder would predominantly have other disfluencies, while subjects with stuttering and with stuttering and phonological disorder would predominantly have the stuttering-like disfluencies.

It is believed that the details of manifestations regarding disfluencies showed by individuals with isolated disorders, on the one hand, and others with disorders in comorbidity, on the other hand, may assist in the differential diagnosis between the conditions and, consequently, favor the best therapeutic conduct.

Assuming that subjects who stutter and subjects with phonological disorder could show differences both regarding

the frequency of occurrence and the typology of disfluencies, this study aimed to identify indicators of speech fluency among subjects who stutter, with phonological disorder and those with the two disorders in comorbidity.

METHODS

Ethical considerations

This is a cross-sectional and prospective observational research with comparison between groups. This study was approved by the Research Ethics Committee (n° 2.070.227). It is noteworthy that all the requirements governing the Resolution of the National Health Council - No. 466/2012 were respected, and the subjects had their participation authorized by signing the Informed Consent Term.

Participants

Data collection to compose the sample occurred during the first semester of 2017. The sample consisted of 30 subjects, both genders, aged 4 to 11 years and 11 months, coming from the Acoustic Analysis Laboratory – (Laboratório de Análise Acústica) LAAC and Fluency Studies Laboratory – (Laboratório de Estudos da Fluência) LAEF, Department of Speech, Language and Hearing Sciences, Faculty of Philosophy and Science, São Paulo State University “Júlio de Mesquita Filho” - UNESP - Marília. Participants were divided into three groups composed of ten subjects each: group with developmental stuttering (SG); group with the diagnosis of phonological disorder (PDG); and group with the two diagnoses in comorbidity (SPDG).

Inclusion criteria for this study were monolingual native speakers of Brazilian Portuguese, without previous speech therapy, aged 4 to 11 years and 11 months. The mean age of SG was 8.40; of the PDG was 6.70; and at SPDG was 6.10 years.

For SG, those who met the diagnostic criterion for persistent developmental stuttering with at least 3% of stuttering-like disfluencies^(9,10) and 12 months of duration of the disfluencies were included.

Specifically, for the PDG, after phonological evaluation, those diagnosed with phonological disorder and who showed spontaneous speech with less than 3% of stuttering-like disfluencies were included; therefore, they did not meet the diagnostic criteria for stuttering.

The diagnosis of phonological disorder was based on the analysis of phonological processes, according to the age at which they should be suppressed, that is, the presence of persistent phonological processes, no longer expected for the child's age group, associated with complementary diagnostic tests, such as: speech perception evaluation, oral diadochokinesia test, multisyllabic word repetition, lexical accent test, among others.

The third and last group included subjects diagnosed with persistent developmental stuttering in comorbidity with phonological disorder.

Those who had neurological, auditory, behavioral, learning disorders, mental disability, genetic syndromes, psychiatric conditions or other relevant alterations that could influence the manifestations of disfluencies or those who, after the collection

and transcription of the speech sample did not have 200 fluent syllables, were excluded from this research. Data regarding various possibilities of alterations that could influence the final diagnosis were obtained by analyzing the medical records of each participant, at the time of admission to the service provided by the School Clinic, were screened and respectively diagnosed.

Procedures

The research procedures were distributed in two stages: speech fluency evaluation and phonology evaluation.

For the speech fluency evaluation, conducted with all sample subjects, an analysis of the spontaneous speech sample was chosen, which constitutes of a greater complexity task for both motor and linguistic⁽¹¹⁾, as well as favors the occurrence of disfluencies in speech when compared to singing, reading, directed speech, among others⁽¹²⁾.

For data collection, digital audiovisual recordings were made of each subject, in interaction with the evaluator. Speech samples were analyzed and transcribed in a total of 200 fluent syllables. Disfluencies were characterized into Stuttering-like Disfluencies (monosyllabic word repetition, syllable repetition, sound repetition, block, prolongation, pause, intrusion) and Other Disfluencies (interjection, hesitation, revision, unfinished words, phrase repetition, non-monosyllabic word repetition), as proposed in the literature⁽¹³⁾.

To determine the frequency of disruptions, the following measures were used: Stuttering-Like Disfluencies (SLD), Other Disfluencies (OD), and Total of Disfluency (TD). To calculate the percentage of SLD, the total number of SLD events was multiplied by 100 and divided by 200, that is, the total of fluent syllables. The same calculations were performed with the total of OD and total of disfluencies (TD, that is, the sum of the SLD and the OD).

For phonological evaluation of PDG isolated or in comorbidity with stuttering, the Speech Evaluation Instrument for Acoustical Analysis (Instrumento de Avaliação de Fala para Análise Acústica - IAFAC) was applied⁽¹⁴⁾. This instrument contains 96 figures, which encompasses the production of words containing all phonemes of the Brazilian Portuguese phonological system, in the context of the vowels /i, a, u/, regarding both the position of simple attack, complex attack, and the position of coda syllable. The data collected through the IAFAC instrument were recorded and later phonetically transcribed.

Statistical analysis

A descriptive and inferential statistical analysis of the data was conducted using STATISTICA 7.0 software. In the comparison between groups, the one-way ANOVA parametric test was used, adopting as independent variable the three groups of subjects (SG, PDG and SPDG), and as dependent variables, the total of Stuttering-Like Disfluencies (SLD) and the total of other types of disfluencies (OD). In cases where there was a significant effect for the group, the Bonferroni post-hoc test was used to verify which groups differed.

When comparing the groups, considering each variable analyzed separately (stuttering-like disfluencies: monosyllabic

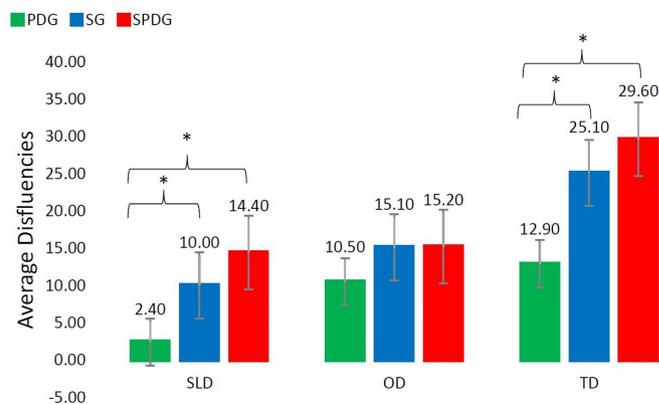
word repetition, part of the word repetition, sound repetition, prolongations, blocks, pauses and intrusions; and other disfluencies: hesitation, interjection, revision, segment repetition, phrase repetition, non-monosyllabic word repetition and unfinished word), the Kruskal-Wallis nonparametric test was used for multiple comparisons.

RESULTS

When comparing the frequency of Stuttering-Like Disfluencies (SLD) and Other Disfluencies (OD) among the three groups studied, Anova One-Way showed a statistical difference between the groups for SLD ($F(2,27) = 14.83, p < 0.00$) and total of disfluencies (TD) ($F(2,27) = 9.74, p < 0.00$), not for total of OD ($F(2,27) = 1.98, p = 0.15$) (Figure 1). Then, a post-hoc analysis was performed using the Bonferroni test to verify which groups differed. It was found that the PDG subjects had a lower frequency of occurrence of SLD and TD compared to SG and SPDG groups ($p < 0.00$), but there was no difference between SG and SPDG groups ($p = 0.17$ for SLD and $p = 0.78$ for TD).

Regarding the typology of disfluencies, it was observed (Table 1), difference of PDG and SPDG in the following variables: “monosyllabic word repetition” ($H(2,30) = 11.73, p < 0.00$); “part of the word repetition” ($H(2,30) = 8.70, p < 0.01$); and “prolongation” ($H(2,30) = 8.98, p < 0.01$). Regarding SLD, for the “block” variable, the PDG is different from both SG and SPDG, ($H(2,30) = 12.63, p < 0.00$), showing a lower frequency of occurrence.

The comparison of the frequency of occurrence of the typologies for other disfluencies as a function of groups is shown in Table 2. Regarding the typologies analyzed in the OD (hesitation, interjection, revision, segment repetition, phrase repetition, non-monosyllabic word repetition and unfinished word), only the variable “interjection” differentiated the PDG from the SG ($H(2,30) = 12.22, p = 0.02$).



Caption: SLD = Stuttering-Like Disfluencies; OD = Other Disfluencies; TD = Total of Disfluencies; SG = developmental stuttering group; PDG = phonological disorder group; SPDG = developmental stuttering and in comorbidity with phonological disorder group; Average Disfluencies

Figure 1. Graph of comparison by pairs for the frequency of Stuttering-Like Disfluencies, Other Disfluencies and Total of Disfluencies. Note: Keys with the asterisks represent the groups that differed from each other. Mean +/- standard error of disfluency event averages of participating groups. *Statistical difference ($p < 0.05$) - Bonferroni test. Source: Created by the author

Table 1. Comparison by typology of Stuttering-Like Disfluencies as a function of groups

Group	Stuttering-like disfluencies						
	MWR	PWR	SR	P	B	PA	I
SG	14.20	17.15	19.55	16.80	19.80	15.95	16.50
PDG	9.55	9.55	11.30	9.55	8.00	12.70	16.50
SPDG	22.75	19.80	15.65	20.15	18.70	17.85	13.50
H value	11.73	8.70	5.99	8.98	12.63	2.40	1.44
P value	0.00*	0.01*	0.05	0.01*	0.00*	0.30	0.48

Caption: SG = developmental stuttering group; PDG = phonological disorder group; SPDG = developmental stuttering and in comorbidity with phonological disorder group; MWR = monosyllabic word repetition; PWR = part of the word repetition; SR = sound repetition; P = prolongation; B = block; PA = pause; I = intrusion; H value = value referring to the distribution of curve H; P value = probability value; * = statistically significant. Source: Created by the author

Table 2. Comparison of the frequency of typologies of other disfluencies as a function of groups.

Group	Typology of other disfluencies						
	H	I	Rev	SegR	PR	NWMR	UW
SG	14.05	22.10	18.65	11.80	16.05	11.85	14.40
PDG	14.40	8.50	16.55	14.50	14.50	14.35	17.70
SPDG	18.05	15.90	11.30	20.20	15.95	20.30	14.40
H value	1.28	12.22	4.11	5.22	1.03	5.65	1.11
P value	0.52	0.02*	0.12	0.07	0.59	0.05	0.57

Caption: SG = developmental stuttering group; PDG = phonological disorder group; SPDG = developmental stuttering and in comorbidity with phonological disorder group; H = hesitation; I = interjection; Rev = revision; SegR = segment repetition; PR = phrase repetition; NMWR = non-monosyllabic word repetition; UW = unfinished words; P value = probability value; H value = value referring to the distribution of curve H. Source: Created by the author

DISCUSSION

This study analyzed and compared the frequency and typology of disfluencies in subjects with stuttering, phonological disorder and with both disorders in comorbidity.

The first hypothesis raised was partially confirmed, regarding that the SG and SPDG showed a higher occurrence of stuttering-like disfluencies when compared to the PDG subjects, however the groups were similar regarding the other disfluencies.

The lower frequency of occurrence of SLD in PDG seems to be related mainly to the fluency profile of fluent subjects, since the stuttering-like disfluencies are uncommon in fluent subjects⁽¹⁵⁾ and do not characterize the manifestations showed by subjects with the phonological disorder.

Subjects from SG and SPDG showed similarities as to the frequency of occurrence of SLD and OD. This finding confirms that SLD characterize the manifestations of stuttering⁽¹⁶⁻²⁰⁾, regardless it is or not associated with phonological disorder. Regarding this result, although SPDG is composed of subjects with disorders in comorbidity, as the quantitative profile of SLD and OD, the group was similar to that found in SG.

This finding corroborates literature descriptions that the excess of stuttering-like disfluencies is the main characteristic of the disorder⁽¹⁶⁻²⁰⁾, and these disruptions constitute one of the most important parameters for diagnosis of stuttering.

Given that speech is the main form of human communication and that its efficiency in transferring information depends on fluency⁽²¹⁾, subjects with stuttering or with stuttering and phonological disorder in comorbidity show significant impairments in communication. It should be emphasized that stuttering is not a simple speech difficulty, but a serious communication problem⁽²²⁾.

Therefore, this finding, added to the fact that 16 to 30% of children who stutter also have phonological disorders^(6,7), should be considered by the speech therapist in the diagnostic process and, consequently, for the planning of a personalized therapy that meets the real needs of each patient. A subject with impaired intelligibility, which may occur due to phonological disorder⁽²³⁾, added to the involuntary stuttering-like disfluencies that occur in the flow of speech, will show greater communication impairment and be more susceptible to bullying. As described in the literature⁽³⁾, this may be an important subgroup of persons who stutter that deserve better attention from researchers.

The analysis and quantitative comparison of the other disfluencies showed that the three groups were similar. It is worth emphasizing that, although the subjects in the three groups have some type of communication disorder, there was no increase in the amount of other disfluencies in relation to the subjects with typical development, based on the reference values indicated in a previous study⁽¹⁵⁾. These findings are consistent with the results of a study⁽²⁴⁾ in which the authors found similarity in the amount of OD between a group with phonological disorder and a control group.

Regarding the language processing, other disfluencies reflect the uncertainties and linguistic inaccuracies, aiming to broaden the comprehension of the message⁽²⁵⁾, justifying the results of this research. These disfluencies are constitutive of the speech dynamics of any person, since speech planning and execution are complex and involve the connection of many brain areas. They may also indicate a way to alleviate speech difficulties at the moment when subjects with phonological disorders try to reach the target pronunciation^(8,24) or subjects who stutter try to avoid SLD.

The second hypothesis was also partially confirmed, since only certain typologies of the SLD and OD showed significant

differences between the groups. In terms of the analyzed typologies of Other Disfluencies, only “interjection” occurred more frequently in SG than in PDG.

Regarding the four typologies of SLD (monosyllabic word repetition, part of the word repetition, prolongation and block), they were less frequent in PDG when compared to SPDG. It seems coherent to explain that the difference found between the groups may be a result of motor complexity during spontaneous speech emission of the SG. In this population, disruptions can be explained by the difference between the generated motor command and the desired motor command during speech, since the greater the motor and melodic complexity during a task, the greater the impairment in speech fluency⁽¹¹⁾.

Regarding the non-differentiation between SG and SPDG for the variables “ monosyllabic word repetition”, “ part of the word repetition” and “prolongation”, it is assumed the presence of dysfunction in the basal ganglia motor circuits, characteristic of the diagnosis of stuttering. Although SPDG concomitantly has the diagnosis of phonological disorder in subjects with stuttering, there is a disorder in the medial system⁽¹²⁾, justifying the occurrence of disfluencies and its non-differentiation between the aforesaid groups, since the task chosen for this research was spontaneous speech, in which the dominance of the medial system occurs mainly when speech conveys thoughts or emotions (present in the speech sample, task of this research).

The non-differentiation of these typologies between SG and PDG may have occurred due to a tendency of smaller occurrence of these disfluencies in SG in relation to SPDG. Although the difference is not statistically significant, it is possible to notice that the mean SLD and total of disfluencies (TD) were higher in SPDG when compared to SG.

With regard specifically to the variable “block”, the subjects of the PDG had a lower frequency of occurrence of this typology, both SG and SPDG. Block is a very characteristic typology of the people who stutter’s speech, is often present in the description of the disorder^(26,27) and its presence is very rare in the speech of people who do not manifest stuttering.

Taking into consideration the experiences, feelings and negative attitudes that subjects with communication disorders may show, more specifically subjects who stutter, it is interpreted that one of the possibilities of higher occurrence of “blocks” in SG and SPDG may result from this freezing response transferred to the speech of these subjects who experience negative emotions perceived as threatening and manifesting as a “freezing response” characterized by an inhibition of movement⁽²⁸⁾.

With regard to the results obtained, it seems reasonable to suspect that both speech disorders - phonological disorder and developmental stuttering - may share underlying aspects, as proposed in a previous study⁽²⁹⁾.

The first shared aspect refers to the fact that both disorders constitute “communication disorders”. Subjects who have some “communication disorder” may acquire a sense of “failure” as a speaker and consequently, may suffer in their attempts at speech production. The second shared aspect concerns a common predisposition to the phonological disorder and developmental stuttering, which could cause similar manifestations in these conditions. Lastly, it is suspected that a third shared aspect by

both disorders would be the presence of a central neurological processing deficit or even a neuromotor disorder or delay, which would lead to interruptions in temporal programming⁽³⁰⁾, providing similar manifestations in the subjects of both groups.

In scientific terms, new study designs may be proposed that contemplate both the analysis of stuttering severity and the analysis of phonological disorder severity. Regarding the clinical implications, it is believed that the speech therapist necessarily needs to analyze the typology of disfluencies to distinguish subjects diagnosed with stuttering, phonological disorder and the two disorders in comorbidity. Moreover, the presence of block represents an important marker for the diagnosis of stuttering.

CONCLUSION

Comparison of the frequency of disfluencies showed that the PDG showed less amount of stuttering-like disfluencies and total disfluencies in relation to SG and SPDG. monosyllabic word repetition, part of the word repetition, and prolongations occurred more frequently in SPDG compared to PDG. Block was less frequent in PDG compared to SG and SPDG.

The three groups were similar in other disfluencies. Interjections were more frequent in SG when compared to PDG.

Since the phonological disorder shows all types of stuttering-like disfluencies, the typology of disruptions seems poorly appropriate as an indicator for the differential diagnosis among the studied conditions, being the frequency with these typologies occur is the best indicator for the differential diagnosis.

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Author contributions

PBAA was responsible for project design and collection, sample selection and case diagnosis, data tabulation and analysis, and manuscript writing; TAP was responsible for data collection and analysis and writing of the scientific article; LMS was responsible for collection, sample selection, data analysis and writing of the scientific article; CMCO was responsible for the study design, data analysis, research co-supervision and article writing, and; LCB was responsible for the project, study design, discussion of the findings and general supervision of the execution stages and manuscript elaboration.