

# SPEECH FLUENCY PROFILE: COMPARATIVE ANALYSIS BETWEEN THE SPORADIC AND FAMILIAL PERSISTENT DEVELOPMENTAL STUTTERING

## *Perfil da fluência: análise comparativa entre gagueira desenvolvimental persistente familiar e isolada*

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### ABSTRACT

**Purpose:** to evaluate and compare the fluency between the familial and the sporadic persistent developmental stuttering, characterizing the typology and the frequency of the disfluencies, the speech rate and the severity of the stuttering. **Methods:** 40 participants aged from 6 to 42 years old, divided in two groups with twenty participants in each one: Familial Persistent Developmental Stuttering and Sporadic Persistent Developmental Stuttering. The procedures used were: clinical and familial history, assessment of fluency and Stuttering Severity Instrument. **Results:** there were no statistically significant differences between the groups regarding the frequency of stuttering like disfluencies, the flow of syllables and words per minute and the severity of stuttering. It was noted a tendency of the group with familial stuttering to show a bigger variability of the severity of stuttering, going from mild to very severe, whereas in the group with sporadic stuttering, the severity varied from mild to severe. **Conclusion:** this study represents the first effort to the characterization of the speech fluency profile of the subgroups of people who stutter, namely familial persistent developmental stuttering and sporadic persistent developmental stuttering. It is possible to conclude that the speech fluency profile of people who stutter, independently of the familial history, is similar. It is noteworthy that the occurrence of some stuttering-like disfluencies, monosyllabic word repetition, block and intrusion were different between groups.

**KEYWORDS:** Speech, Language and Hearing Sciences; Speech; Stuttering; Speech Disorders; Genetics

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### ■ INTRODUCTION

Developmental stuttering is a communication disorder that is typically characterized by excessive and/or lengthy breaks in fluency during linguistically formulated speech <sup>1</sup>.

The main manifestation of stuttering is the intermittent failure of the nervous system to generate appropriate command signals to the muscles whose activity must be dynamically controlled for fluent speech to be produced <sup>2</sup>.

The etiology of stuttering is still unknown, but there is a consensus that genetic factors play a role in about half of all cases of persistent developmental

stuttering<sup>3</sup>. This stuttering subgroup was named Familial Persistent Developmental Stuttering<sup>3,4</sup>. The other stuttering subgroup, which originates in childhood without genetic predisposition is known as a Sporadic Persistent Developmental Stuttering.

The main arguments supporting the genetic factors of stuttering include: 1) higher concordance between monozygotic twins (62.5% to 90%) than dizygotic twins (6.6% to 9%)<sup>5</sup>; 2) stuttering is more likely to develop among consanguineous individuals than in cases where the affected individuals do not have such relationship<sup>5</sup>; 3) the similarity of the phenotypic trait developed between stutterers such as repetitions, prolongations of sounds or word syllables not associated to differences in language and culture<sup>4</sup>.

However, contemporary theories related to stuttering reveal that the disorder is multifactorial, so no single constitutional or environmental factor is sufficient to justify the clinical condition<sup>6</sup>. It is also known that the spectrum of risk factors related to stuttering is broad and heterogeneous<sup>7</sup>. Therefore, this disorder results from a complex influence of multiple factors including genetic predisposition, motor skills for speech, linguistic, cognitive, emotional and environmental factors<sup>8</sup>. For scholars<sup>8</sup>, the importance of each factor and how they interact with other factors over time probably result in considerable differences among individuals who stutter. As an environmental factor, the literature highlights the emotional factor, which can affect the development of the disorder or its severity through personal factors and family conflicts<sup>9</sup>.

Among the biological factors are highlighted those with higher significance for the development of the disorder: prenatal, medical, developmental, language and genetic history<sup>5,10</sup>.

The stuttering risk factors were studied in both subgroups, familial and sporadic, and the results showed that they were similar, regardless of family history<sup>11</sup>. The most prominent factors in both groups were: male gender; the persistence of the disorder, with disfluencies that last more than 12 months; the presence of stuttering-like disfluencies and qualitative factors associated with the disfluencies, and negative family attitude and personal reaction<sup>11</sup>.

When comparing the risk factors in both groups, only the stressors close to the emergence of disfluencies presented statistically significant difference. Therefore, the authors believe that cases without positive family history for stuttering require additional risk factors, reason why the sporadic developmental stuttering group may have presented a significantly higher number of stressors in relation to the familial stuttering group<sup>11</sup>.

A recent study on the analysis of risk factors in cases of sporadic stuttering revealed that the risk increases when disfluencies last longer than 12 months and when there are associated qualitative and communicative factors<sup>12</sup>.

However, the most widely known communication function measures for individuals who stutter are quantitative (such as the frequency of stuttering-like disfluencies) and qualitative (such as disfluency types)<sup>13</sup>. Furthermore, another measure used and recommended by the literature is the speech rate, which indicates the communicative productivity<sup>13</sup>.

Therefore, the purpose of the study was to evaluate and to compare the fluency between the unrelated individuals with familial persistent developmental stuttering and those with sporadic persistent developmental stuttering, characterizing the typology and the frequency of the disfluencies, the speech rate and the stuttering severity.

## METHODS

This research study comprises an experimental and cross-sectional comparison between groups, conducted with individuals who stutter of the *Laboratório de Estudos da Fluência – LAEF* of the *Centro de Estudos da Educação e da Saúde* of Universidade Estadual Paulista – FFC – Marília.

The study included 40 subjects (33 males and 7 females) aged between 6 and 42 years old (mean age of 14.7 years, SD = 9.69). The participants were divided into two groups: patients with Familial Persistent Developmental Stuttering, titled PWFS (People With Familial Stuttering) composed of 20 unrelated individuals (belonging to different families) aged between 8 and 42 years old (mean age of 18.75 years, SD = 10.42) from both genders (17 males and 3 females), and patients with Sporadic Persistent Developmental Stuttering, titled PWSS (People With Sporadic Stuttering) composed of 20 individuals aged between 6 and 28 years old, from both genders (16 males and 4 females).

The inclusion criteria were that participants had to be native speakers of Brazilian Portuguese; be over six years old; have started stuttering during childhood (developmental); the stuttering condition had to have persisted for more than one year without remission (persistent); present at least 3% of stuttering-like disfluencies, and present at least mild stuttering according to the Stuttering Severity Instrument – SSI<sup>14</sup>.

Individuals included in the Familial Persistent Developmental Stuttering (PWFS) group presented a positive family history for stuttering (familial), that is, had at least one relative who stuttered. Individuals included in the Sporadic Persistent Developmental

Stuttering (PWSS) group did not have a positive family history for stuttering.

The exclusion criteria for participants were: present any neurological disorder in the family, whether genetic or not, such as dystonia, extra pyramidal diseases, mental disorder, epilepsy, attention deficit hyperactivity disorder (ADHD), psychiatric symptoms or conditions; present oral communication alterations that are not compatible with their age; present conductive or sensorineural hearing loss; and other pertinent conditions that could cause diagnostic errors.

The parties responsible for all children participating in the study, or the participants themselves (if over 18 years old) have agreed in writing to their participation, based on the clarifications contained in the Informed Consent form that was presented to them.

All study participants underwent the following procedures:

- **clinical history:** conducted with the parents or the individuals themselves (adults), in order to obtain data regarding the age of stuttering onset (to determine if it was developmental) , among others.
- **family history:** the pedigree was constructed based on information received from the participants and/or their parents in order to separate the participants between the familial or sporadic groups.
- **speech fluency evaluation:** subjects were videotaped in order to obtain speech samples containing 200 fluent syllables for analysis and comparison of the results. After collecting the participants' speech samples, these were transcribed, considering the fluent and non-fluent syllables. This was followed by an analysis of the speech sample and the types of disfluencies were characterized accordance to the following description: *other disfluencies:* hesitations, interjections, revisions, unfinished words, non-monosyllabic word repetitions, repetition of segments and phrases; *stuttering-like disfluencies:* monosyllabic word repetitions, sound repetitions, syllable repetitions, prolongations, blocks, pauses and intrusions. The following measures we used to characterize

the frequency of the disruptions: percentage of speech discontinuity or speech disruption rate, and percentage of stuttering-like disfluencies or stuttering disruption rate. The speech rate was measured according to the test used, characterizing the flow of syllables and words per minute<sup>15</sup>.

- **Stuttering Severity Instrument (SSI-3)**<sup>14</sup>: used for each participant, classifying their stuttering as mild, moderate, severe or very severe. This test assessed the frequency and duration of stuttering-like disfluencies, as well as the presence of physical concomitants associated with the disfluencies, according to the protocol proposed by Riley.

Once the evaluation was completed, the family received the feedback of the test results. Information about stuttering was offered through guidance and instructions in an informational report, and the cases were referred for speech therapy at the same Center where the project was developed.

This study was approved by the originating institution's Research Ethics Committee under protocol No. 0091 /2011.

The Mann- Whitney test was used for statistical analysis in order to verify possible differences between the groups. Another statistical method used was the application of the Likelihood Ratio test in order to verify possible differences between groups regarding stuttering severity, a variable of interest for the study. The significance level adopted for the application of statistical tests was 5% (0,050). Data analysis was conducted using the SPSS (Statistical Package for Social Sciences) software, version 19.0.

## ■ RESULTS

According to the purpose of the study, the results obtained are presented in the following Tables. By comparing the groups it is observed that the PWFS and PWSS groups did not differ statistically in three of the four measures examined, namely stuttering-like disfluencies, flow of syllables per minute and flow of words per minute (Table 1).

**Table 1 – Comparison between groups regarding the occurrence of speech discontinuity, stuttering-like disfluencies, flow of syllables and words per minute**

Groups	PWFS				PWSS				p-value
	Mean	SD	Minimum	Maximum	Mean	SD	Minimum	Maximum	
Speech discontinuity	19.10	13.98	9	62.50	10.89	3.15	6.9	19.50	<b>0.001*</b>
Stuttering-like disfluencies	8.93	10.01	3.00	45.00	5.56	2.03	3.00	11.00	0.860
Flow of syllables per minute	177.88	68.27	35.71	266.67	168.56	38.86	94.50	238.80	0.292
Flow of words per minute	99.86	38.54	21.25	150.61	108.19	23.71	78.46	160.00	0.922

Note: PWFS = Group of People With Familial Stuttering; PWSS = Group of People With Sporadic Stuttering; SD = standard deviation.  
 \*\*Statistical significance ( $p \leq 0.05$ ) – Mann-Whitney Test – significant values are shown in bold and with an asterisk

The other disfluencies in the groups of participants are distributed by typology in Table 2. For both groups, the most frequent typology was hesitation, and the least frequent typology was segment repetition. Most other disfluencies (revisions, unfinished words, sentence and segment repetition) presented quantitative similarities between the

participating groups. However, the PWFS group presented a higher incidence with statistically significant difference compared to the PWSS group for the following other disfluencies typologies: word repetition (5.14 times more frequent), interjection (1.98 times more frequent) and hesitation (1.87 times more frequent).

**Table 2 – Comparison between the groups regarding hesitation, interjection, revision, unfinished word, phrase repetition, segment repetition and word repetition**

	H		I		Rv		UW		PR		SegR		WR	
	M	SD	M	SD	M	SD	M	SD	M	SD	M	SD	M	SD
PWFS	8.05	6.55	5.85	4.27	0.95	0.76	0.30	0.57	1.45	1.91	0.15	0.37	3.60	2.37
PWSS	4.30	4.85	2.95	3.33	0.80	1.28	0.25	0.55	0.80	1.11	0.25	0.72	0.70	0.92
p-value	<b>0.016*</b>		<b>0.006*</b>		0.144		0.724		0.192		0.948		< <b>0.001*</b>	

Note: PWFS = Group of People With Familial Stuttering; PWSS = Group of People With Sporadic Stuttering; M = mean; SD = standard deviation; H = hesitation; I = interjection; Rv = revision; UW = unfinished word; PR = phrase repetition; SegR = segment repetition; WR = word repetition.

\*\*Statistical significance ( $p \leq 0.05$ ) – Mann-Whitney Test – significant values are shown in bold and with an asterisk

In the analysis of the occurrence of stuttering-like disfluencies in the participating groups, it can be observed that the number of repetitions of part of the word, sound repetitions, prolongations and pauses is similar (Table 3). However, there was a statistically significant differences related to monosyllabic word repetition, blocks and intrusions, where the PWSS group presented 5.35 times more monosyllabic word repetitions than the PWFS group. The greater occurrence of blocks and intrusions was presented by the PWFS group (5 times more blocks and 6.66 times more intrusions). In the PWFS group, the

most common stuttering-like disfluencies was block, and for the PWSS group it was monosyllabic word repetition. The least frequent stuttering-like disfluencies presented by the PWFS group was pause, and by the PWSS group was intrusion.

(1.86 times more interjections, 4.91 times more revisions and 12.28 times more unfinished words). For the PCT group, the typology with greater

As for the stuttering severity, the groups did not present statistically significant differences ( $p = 0.145$ ) (Table 4). Note that both groups presented a greater number of mild stuttering.

**Table 3 – Comparison between groups regarding repetition of monosyllabic words, repetition of part of the word, sound repetition, prolongation, block, break and intrusion**

	RMW		RPW		SR		P		B		Pa		In	
	M	SD	M	SD	M	SD	M	SD	M	SD	M	SD	M	SD
PWFS	1.00	1.72	2.20	2.48	1.60	2.72	4.90	8.52	6.25	6.50	0.90	1.68	1.00	1.38
PWSS	5.35	4.23	1.30	1.72	0.60	1.05	2.05	2.42	1.25	2.12	0.55	0.95	0.15	0.37
p-value	<b>&lt; 0.001*</b>		0.118		0.101		0.145		<b>&lt; 0.001*</b>		0.638		<b>0.021*</b>	

Note: PWFS = Group of People With Familial Stuttering; PWSS = Group of People With Sporadic Stuttering; M = mean; SD = standard deviation; RMW = repetition of monosyllabic word; RPW = repetition of part of the word; SR = sound repetition; P = prolongation; B = block; Pa = pause; In = intrusion.

\*\*Statistical significance ( $p \leq 0.05$ ) – Mann-Whitney Test – significant values are shown in bold and with an asterisk

**Table 4 – Comparison between the groups regarding the stuttering severity in the Stuttering Severity Instrument (Riley, 1994)**

	Stuttering Severity				Total
	Mild	Moderate	Severe	Very severe	
PWFS	14 70%	3 15%	2 10%	1 5%	20 100%
PWSS	10 50%	3 15%	7 35%	0 0%	20 100%
Total	24 60%	6 15%	9 22,5%	1 2,5%	40 100%
p-value	0.145				

Note: PWFS = Group of People With Familial Stuttering; PWSS = Group of People With Sporadic Stuttering.

\*\*Statistical significance ( $p \leq 0.05$ ) – Likelihood Ratio test

## ■ DISCUSSION

There is currently a trend in stuttering research in analyzing different subgroups<sup>11,12,16,17</sup> in an attempt to improve the knowledge about them in order to improve the diagnosis and therapeutic intervention of the disorder. The familial developmental stuttering subgroup has received increased attention from researchers, due to a growing interest in the search for candidate genes for transmission of the disorder<sup>18-22</sup>. However, few investigations<sup>12</sup> have devoted to analyzing the sporadic stuttering subgroup. Therefore, the purpose of this study was to evaluate and compare the fluency between the unrelated individuals with Familial Persistent Developmental Stuttering (PWFS) and those with Sporadic Persistent Developmental Stuttering (PWSS), characterizing the typology and the frequency of the disfluencies, the speech rate and the stuttering severity.

Regarding to the age of the participants, there was a statistically significant difference between the groups ( $p < 0.001$ ). However, an earlier investigation

conducted with the same stuttering subgroups (familial and sporadic) with an infant population showed no difference with respect to age<sup>11</sup>.

Disfluencies are part of language production, as they help the speaker to produce a more appropriate speech, both in content and in form<sup>23</sup>. As described in the literature<sup>24,25</sup>, other disfluencies mainly reflect the linguistic uncertainties and inaccuracies, and are considered common to all speakers.

The findings of this research revealed that quantitatively the PWFS and PWSS groups only showed statistically significant differences related to the total number of speech disruptions (other and stuttering-like disfluencies). A possible explanation for this finding is that since the PWFS group has a greater chronological age, its participants can use more other disfluencies in an attempt to delay or prevent stuttering-like disfluencies. It is known that the longer the time an individual lives with stuttering, the greater the possibility of using delay or avoidance resources.

As for the qualitative analysis of other disfluencies, data revealed that the groups were similar

for most of them (revision, unfinished word, phrase repetition and segment repetition). Thus, the findings lead us to believe that regardless of the stuttering subgroup (familial or sporadic), people who stutter have a similar qualitative profile with respect to other disfluencies.

However, some typologies (word repetition, interjection and hesitation) were more frequent in the PWFS group. When analyzing this finding it was found that there is no relationship with stuttering severity, considering that even though the groups did not present statistically significant difference related to severity, the PWSS group presented a tendency to have more cases of severe stuttering than the PWFS group. Another hypothesis that could explain this finding is the age of the participants, since other disfluencies indicate that the speaker is looking for the solution<sup>26</sup>, and hesitation pauses and interjections indicate that the speaker has perceived a problem before producing speech<sup>27</sup>. Therefore, it can be considered that the higher chronological age, or the longer the duration of disfluencies, the greater is the perception of stuttering-like disfluencies, and therefore the person who stutters could use these other disfluencies in an attempt to prevent the onset of stuttering. We emphasize the fact that the PWFS group had a greater chronological age than the PWSS group.

Stuttering-like disfluencies are typical of people who stutter<sup>28,29</sup>; however, it can also occur in a lesser amount in the speech of fluent persons<sup>24,30</sup>. These disfluencies seem to indicate a rupture in the speech related to its motor execution<sup>31</sup>. The results of this research showed that the PWFS and the PWSS groups showed no statistically significant quantitative difference. The compiled literature did not present any studies related to fluency profile in these subgroups, which hinders the discussion of these findings. It is believed that the quantitative profile of stuttering-like disfluencies of the PWFS and PWSS subgroups are similar, that is, independent of their etiological nature.

The results of the qualitative analysis of stuttering-like disfluencies showed that the groups were similar in 57.14% of the typologies (repetition of part of the word, sound repetition, prolongation and pause). However, the groups differed regarding monosyllabic word repetition, block and intrusion, where the PWFS group presented a higher frequency of block and intrusion.

Note that stuttering is a disorder with a very high variability characteristic<sup>32</sup>. Thus, the results of this research support the literature regarding the variability of stuttering, and lead to believe that the

quantitative and qualitative profiles of the PWFS and PWSS subgroups are similar.

As described in the literature<sup>33</sup>, the flow of syllables and words per minute in people who stutter are reduced if compared to people who do not stutter. However, regardless of etiology, that is, familial or sporadic, the participating groups were similar in regard to the flow of syllables per minute, also called articulatory rate, and also in regard to the flow of words per minute, or production of information.

This decrease in the speech rate of people who stutter is possibly related to the increase in the amount of disfluencies in the speech flow<sup>33</sup>. We can observe that the minimum flow of syllables per minute (35.71 SPM) and words per minute (21.25 WPM) in the PWFS group were significantly reduced compared to the values found in the PWSS group (94.50 SPM and 78.46 WPM). These findings can be explained by the fact that one of the participants in the PWFS group presented a very severe stuttering, with 45% of stuttering-like disfluencies and 17.5% of other disfluencies, whereas in the PWSS group the maximum values were 11% of stuttering-like disfluencies and 11.5% of other disfluencies.

Based on data obtained from stuttering severity, it can be observed that there was no statistically significant difference between the PWFS and PWSS groups included in this study. However, note that the PWFS group showed a tendency to present greater variability of stuttering severity, which was from mild to very severe. In the PWSS group the severity ranged from mild to severe. Another observation with respect to severity is the tendency shown by the PWFS group of having more people with mild stuttering (70%) compared to the PWSS group (50%). In the PWSS group 35% of participants expressed severe stuttering, while in the PWFS group 10% of participants were classified as having severe stuttering, and 5% as having very severe stuttering.

## ■ CONCLUSION

This study represents the first effort to the characterization of the speech fluency profile of the subgroups of persons who stutter, namely familial persistent developmental stuttering and sporadic persistent developmental stuttering. Based on the analysis of the data obtained it can be concluded that regardless of family history, the speech fluency profiles among people who stutter are similar concerning percentage of stuttering-like disfluencies, flow of syllables and words per minute, and stuttering severity.

It is noteworthy that the occurrence of some stuttering-like disfluencies, monosyllabic word repetition, block and intrusion were different between groups. However, due to sample size these data cannot be generalized, and it is believed that they are related to the variability of stuttering.

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## RESUMO

**Objetivo:** avaliar e comparar a fluência de indivíduos com Gagueira Desenvolvimental Persistente Familiar e de indivíduos com Gagueira Desenvolvimental Persistente Isolada, caracterizando a tipologia das disfluências, a porcentagem de disfluências, a taxa de elocução e a gravidade da gagueira.

**Métodos:** participaram 40 indivíduos, entre 6 e 42 anos de idade, divididos em dois grupos de 20 participantes cada um: Gagueira Desenvolvimental Persistente Familiar e Gagueira Desenvolvimental Persistente Isolada. Os procedimentos utilizados nos grupos foram: histórias clínica e familiar, avaliação da fluência e Instrumento de Gravidade da Gagueira. **Resultados:** não houve diferenças estatisticamente significantes entre os grupos quanto à porcentagem de disfluências gagas, ao fluxo de sílabas e palavras por minuto, e a gravidade da gagueira. Nota-se uma tendência do grupo de pessoa com gagueira familiar apresentar uma maior variabilidade da gravidade da gagueira, que foi de leve a muito grave, enquanto que, no grupo de pessoas com gagueira isolada, a gravidade variou de leve a grave. **Conclusão:** este estudo representa um primeiro esforço para a caracterização do perfil da fluência de subgrupos de pessoas que gaguejam, a saber, gagueira desenvolvimental persistente familiar e gagueira desenvolvimental persistente isolada. Pode-se concluir que o perfil da fluência de pessoas com gagueira, independente do histórico familiar, é semelhante. Vale ressaltar que a ocorrência de algumas tipologias gagas, repetição de palavra monossilábica, bloqueio e intrusão foram distintas entre os grupos.

**DESCRITORES:** Fonoaudiologia; Fala; Gagueira; Distúrbios da Fala; Genética

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