Fluency aspects in the oral narrative of individuals with Fetal Alcohol Spectrum Disorder

Aspectos da fluência na narrativa oral de indivíduos com Transtorno do Espectro Alcoólico Fetal

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ABSTRACT

Purpose: To investigate fluency aspects in the oral narrative of individuals with Fetal Alcohol Spectrum Disorder (FASD), and to compare them with individuals with typical language development regarding frequency of disfluencies and speech rate. **Methods:** Participants were nine individuals with FASD (two with Fetal Alcohol Syndrome and seven with Alcohol-Related Neurodevelopmental Disorder) and chronological ages between 4 and 12 years. This group was matched to nine individuals with typical language development by gender and chronological age. Oral narratives were produced using the book "Frog, where are you?", and analyzed according to the speech fluency parameters of the *Teste de Linguagem Infantil* – ABFW (type of disfluency, frequency of ruptures, and speech rate). **Results:** The FASD and typical language development groups differed on overall frequency of disfluencies, typical disfluencies and stuttering disfluencies. For both groups, the most frequent types of disfluencies were hesitation and pause. **Conclusion:** The results suggest that the increased frequency of pauses and hesitations in individuals with FASD might be due to difficulties in the linguistic elaboration of oral narratives, justifying the lower speech rate showed by these individuals.

Keywords: Fetal Alcohol Syndrome; Language; Speech; Narration; Speech production measurement

RESUMO

Objetivo: Investigar aspectos da fluência na narrativa oral de indivíduos com Transtorno do Espectro Alcoólico Fetal (TEAF), e compará-los a indivíduos com desenvolvimento típico de linguagem quanto à frequência de disfluências e à velocidade de fala. **Métodos:** Participaram deste estudo nove indivíduos com TEAF (dois com Síndrome Alcoólica Fetal e sete com Transtorno Neurodesenvolvimental Relacionado ao Álcool) com idades cronológicas entre 4 e 12 anos de idade, pareados a outros nove indivíduos com desenvolvimento típico de linguagem segundo gênero e idade cronológica. As narrativas orais foram produzidas utilizando o livro *Frog, where are you?* e analisadas quanto aos parâmetros da fluência de fala do Teste de Linguagem Infantil – ABFW (tipologia de disfluências, frequência de rupturas e velocidade de fala). **Resultados:** Os grupos com TEAF e desenvolvimento típico de linguagem diferiram quanto à frequência total de disfluências, disfluências comuns e disfluências gagas, sendo as tipologias mais frequentes a hesitação e a pausa. **Conclusão:** Os resultados sugerem que a frequência aumentada de pausas e hesitações dos indivíduos com TEAF pode ser decorrente de dificuldades na elaboração da narrativa oral de histórias, justificando a menor taxa de velocidade de fala apresentada por esses indivíduos.

Descritores: Síndrome Alcoólica Fetal; Linguagem; Fala; Narração; Medida da produção da fala

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INTRODUCTION

The Fetal Alcohol Syndrome (FAS) was described in 1973 in a study with a group of children born to alcoholic mothers and presenting a series of deficiencies in pre- and/or post--natal development; facial dysmorphism (e.g. undefined labial philtrum, eyelid fissure, fine upper lip, flattened face), and a dysfunction in the Central Nervous System (e.g., intellectual disability and/or attention deficit disorder)[1]. Even though this condition was acknowledged in 1973, these characteristics had already been reported in a 1968[2] study of 127 cases of children exposed to alcohol during the gestational period.

With the advance of research on the effects of alcohol during fetal development, a wide range of clinical signs among individuals has been described, both for the type and the degree of compromise. Thus, in 2000[3] a more comprehensive term was proposed to refer to the set of clinical characteristics resulting of exposure to alcohol during fetal development: Fetal Alcohol Spectrum Disorder (FASD). This term has been widely used in most studies to faithfully represent the extensive and varied spectrum of clinical signs observed in individuals whose mothers presented a positive history of alcohol consumption during pregnancy[4].

Along the years, a wide range of variability of clinical signs has been incorporated into this condition of (teratogenic) environmental etiology, and this has led researchers to propose diagnostic subclassifications based on the FASD continuum considering, in addition to the identified clinical sign, the severity of its manifestation, representing the subphenotypes of this condition. The edges of this continuum are represented by the Fetal Alcohol Syndrome (FAS), presenting the most severe degree of phenotypic compromise, and by the Alcohol-Related Neurodevelopmental Disorder (ARND), presenting the mildest degree of phenotypic compromise[5]. The variety of clinical pictures of individuals with FASD depends mainly on several factors, such as: the fetal developmental period in which the mother consumed alcohol; the number of doses consumed by the mother during pregnancy; the mother's nutritional condition; and the individual susceptibility of the fetus to absorb the alcohol consumed by the mother.

Among the physical and neuroatomic characteristics described in studies on FASD[4-7], the concern to describe the cognitive, emotional, behavioral and social adaptation aspects[4,5], as well as oral language[6,7], seems to be increasingly more frequent.

Two studies on spoken language production have addressed more specifically the change in fluency in children with FASD, but both have chosen verbal fluency tasks with focus on work memory and evocation of words, which were not included in the scope of this study. The first of the previous studies characterized the changes in the executive functions of individuals with FASD, describing losses in abstract thought, planning and cognitive flexibility[8]. The second study addressed the development of 18 children with FASD also in executive functions, including verbal and non-verbal fluency of a group of ten individuals with a FAS diagnosis, a group of eight cases without this diagnosis, but with Fetal Alcohol Spectrum Disorder and a control group made up of ten children with ages ranging from 8 to 15 years. The results of this study suggest alterations in verbal fluency of individuals exposed to alcohol, both with FAS diagnosis and those exposed to alcohol but without the FAS diagnosis. Alterations in verbal fluency were not found in children of the control group. This result confirmed, according to the authors, that the alterations in fluency were common in children with FASD, regardless of the degree of compromise caused by alcohol consumption – FAS or ARND[9].

The compromise of the executive function in the FASD was explained by the authors as the impairment in abstraction concepts, work memory, and speech recognition, deficits in communicative ability, including morphosyntactic, phonological, semantic and pragmatic, mainly in spoken language production[5-7].

Considering that the compiled studies did not focus on the speech fluency of individuals exposed to alcohol during fetal development, the purpose of this study was to investigate fluency aspects in oral narrative of individuals with FASD, diagnosed with FAS or ARND. In this work, the proposal was to analyze the fluency in the oral narrative of a story as for the frequency of disfluencies and speech rate of these individuals, comparing them to individuals with no history of alcohol use during pregnancy and with typical language development.

METHODS

The present research was approved by the Ethical Committee Committee of the School of Philosophy and Sciences of Universidade Estadual Paulista "Júlio de Mesquita Filho" (UNESP) Marília (SP) (process nº 1548/2010) and the participation of individuals was authorized by their parents or guardians, who signed Consent Form.

Nine individuals diagnosed with FASD took part in this study, with chronological age between 4 and 12 years (M=8.0 years old). These subjects presented positive history of maternal alcohol consumption during pregnancy and the particular manifestations of the condition. Two cases were diagnosed as FAS by a specialist, according to the diagnostic criteria of this condition (growth deficiency, facial phenotype, microcephaly and intellectual disability).

The other seven subjects fell under the ARND picture, presenting variability as for the presence of ARND and a milder degree of compromise, such as the presence of some facial features, growth deficiency and varied behavioral problems confirmed by a genetic assessment (Chart 1).

Individuals with FASD were compared to those with typical language development (TLD), paired according to gender and chronological age. The TLD group was constituted following

criterion of negative history of maternal alcohol drinking during pregnancy and absence of neurological, perceptual, sensorial and emotional deficits, as well as speech and language alterations.

Oral narrative production was obtained through the storybook Frog, where are you?[10] made up of 29 boards of sequential events with no written text. At first, the individual was advised to handle the book from the beginning to the end so as to subsequently use this material to the story generation.

All narrative samples were filmed and taped so they could be transcribed later on and analyzed for speech fluency aspects, according to the Teste de Linguagem Infantil – ABFW – Fluency[11], which includes typical disfluencies (hesitation, interjection, revision, unfinished word, word repetition, segment repetition, sentence repetition) and stuttering disfluencies (syllable and/or sound repetition, prolongation, blocking, pause (silent pause) and intrusion of sound and/or segment). Following the said classifications, the ruptures were analyzed for the percentage of occurrence of: (1) each type of typical and stuttering disfluency; (2) typical disfluency, from the total of typical disfluencies; (3) stuttering disfluency from the total of stuttering disfluencies; and (4) total disfluency from the total of disfluencies, represented by the sum of typical and stuttering disfluencies.

In this study, the occurrence of disfluencies was recorded throughout the story oral narrative sample, and the compatibility calculation of the number of fluent syllables was carried out as suggested by the ABFW test, regarding the number of occurrence of typical and stuttering disfluencies for the purposes of comparison between participants. Speech rate was measured by the flow of words and syllables by minute and also following the criterion suggested by the ABFW test, using the total of fluent words and syllables with regard the total time of duration of the narrative.

Data was initially analyzed by comparing fluency aspects of each one of the cases with FASD with the respective TLD group. Subsequently, a descriptive analysis was carried out to verify data dispersion, considering the minimum and maximum numerical value reached by the group with FASD and by the group with TLD. For the purpose of comparison between the groups (FASD and TLD), the Mann-Whitney non-parametric test was applied with 5% significance level. The statistical analysis was carried out using Minitab 16 statistical software.

RESULTS

Table 1 presents the disfluencies identified in oral narration of individuals with FASD and TLD. Out of the seven cases of ARND analyzed, four presented a higher percentage of typical disfluencies than their counterparts by chronological age, and this event was also observed for the two cases with FAS diagnosis. As for the percentage of stuttering disfluencies, all

Chart 1. Clinical characteristics of individuals with Fetal Alcohol Spectrum Disorder (FASD)

Variables	ARND							FAS	
	S1	S2	S3	S4	S5	S6	S7	S8	S9
Gender	M	M	M	F	M	M	M	F	F
Age	4	5	6	7	8	9	11	10	12
Pre-natal exposure to alcohol	+	++	+	++	++	++	++	++	++
Growth deficiency									
Low weight at birth	NE	+	NE	+	NE	NE	NE	+	+
Low height	-	+	-	+	-	-	-	+	+
Facial phenotype									
Small eyelid fissure	++	+	+	++	+	+	+	+++	++
Epicanthic fold	++	-	-	++	+	+	+	+++	++
Lowered nasal bridge	-	+	+	++	-	-	-	+++	++
Absence of nasal filter	++	+	+	+	+	-	-	+++	++
Fine upper lip	++	-	+	+	-	-	+	+++	++
Alterations in the CNS									
Microcephaly	-	-	-	-	-	-	-	+++	++
Structural alterations	NE	NE	NE	NE	NE	NE	NE	NE	NE
Neurodevelopmental alterations									
Behavioral problems	++	+++	++	+++	-	-	-	+++	+++
Intellectual disability	-	NE	NE	NE	-	-	-	+++	++
Hearing impairment	-	NE	-	-	-	-	-	+++	-

Note: ARND = Alcohol-related neurodevelopment disorder; FAS = Fetal Alcohol Syndrome; M = male; F = female; NE = not evaluated; (+) = slight alteration; (++) = moderate alteration; (-) = appropriate; (+++) = severe alteration; CNS = Central Nervous System

cases of alcohol exposure presented values higher than their counterparts by chronological age. This data is confirmed in the statistical analysis with higher median for the FASD group (Table 2).

Typical disfluencies such as sentence repetition and stuttering disfluencies such as syllable repetition, prolongations, blockings and intrusion of sounds or segments were not observed for individuals with ARND, FAS and TLD.

Among the observed disfluencies in oral story narration, hesitation and pause were the ones occurring with the highest frequency, both for individuals with FASD and with TLD; however, the groups presented a significant difference as a result of the higher median presented by the group with FASD (Table 2).

In the analysis of the development of each case with FASD with compared with its counterpart as for speech rate, cases with TLD were found to present numbers of syllables and words per minute higher than those of subjects exposed to alcohol (Table 1), except for a case with ARND (S1), which presented values close to those of its counterpart with the same chronological age. This data may also be seen in Table 2, with lower median for speech rate measures of the group with FASD compared with the group with TLD.

During the story oral narrative task, performance varied between the cases with ARND and FAS, characterized by a reduced lexical repertoire, naming errors and lexical access difficulties. In general, more serious difficulties were observed in wording construction of cases with FAS than in the construction of complex sentences by those with ARND, with significant mistakes in the use of grammar rules and organization of sentence elements.

DISCUSSION

When investigating the aspects of fluency in story oral narrative of individuals with FASD and TLD, of the same gender and similar chronological age, the most frequent disfluencies found were hesitation, followed by pause and revision. However, the percentage of these disfluencies in the narrative was evidently higher for cases exposed to alcohol, mainly for hesitation and pause (Tables 1 and 2).

When comparing the frequency of disfluencies presented by individuals with ARND (S5 and S6) and FAS (S8 and S9) with similar chronological ages (Table 1), we note that the percentage of disfluencies found for cases with FAS is higher than the one presented by individuals with ARND, as it happened for the frequency of stuttering disfluencies. The results suggest that the percentage of individual disfluencies was variable between these individuals, and hesitation was more frequent in the narrative presented by case S8 (FAS) to the detriment of S5 (ARND). In turn, the percentage of hesitation was higher for

Table 1. Percentage of disfluencies and speech rate in oral story narrative by individuals with the Fetal Alcohol Spectrum Disorder and typical language development

	FASD									TLD								
	ARND							FAS										
	S1	S2	S3	S4	S5	S6	S7	S8	S9	C1	C2	C3	C4	C5	C6	C7	C8	C9
Age	4	5	6	7	8	9	11	10	12	4	5	6	7	8	9	11	10	12
Typical disfluencies																		
HES	2.5	2.8	3.3	1.8	2.9	3.6	1.4	4.7	2.4	1.8	0.2	1.0	2.0	0.7	0.0	2.0	0.8	1.4
INT	3.1	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	1.6	0.0	0.0
REV	1.2	1.4	0.6	1.8	1.4	1.2	0.3	0.9	2.0	0.9	0.8	1.0	1.5	0.7	1.1	1.6	0.4	1.1
UW	0.0	0.7	0.0	0.7	0.3	0.0	0.1	0.0	0.2	0.9	0.0	0.5	0.6	0.5	0.3	0.4	0.2	0.3
WR	0.0	1.4	1.7	0.2	0.0	0.0	0.1	0.6	0.6	1.3	0.0	0.1	0.2	0.0	0.9	1.3	0.0	0.8
SeR	0.0	0.2	0.0	0.0	0.0	0.0	0.1	0.0	0.2	0.4	0.0	0.0	0.0	0.2	0.3	0.0	0.0	0.3
% typical disfluencies																		
	7.0	6.6	5.8	3.7	4.8	4.9	2.0	6.1	6.5	5.5	1.1	3.1	2.7	4.5	2.3	2.8	1.6	4.1
Stuttering disfluencies																		
SoR	0.0	0.0	0.0	0.1	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0	0.0
PAU	2.8	3.9	5.6	1.0	3.5	3.5	0.8	1.5	1.1	1.8	1.1	0.5	0.4	0.7	0.3	0.0	0.0	0.0
% stuttering disfluencies																		
	2.8	3.9	5.6	1.4	3.5	5.7	1.0	1.8	1.8	1.8	1.4	2.6	0.0	0.4	0.7	0.3	0.0	0.1
Speech rate																		
WM	62.2	54.2	40.3	118.2	55	54.1	108.2	49	86.3	65.6	83.4	79.6	79.7	100	94.1	67.4	138	140
SM	102	102	70	216	99	95	200	89	155	103	149	151	139	195	167	119	280	242

Note: FASD = Fetal Alcohol Spectrum Disorder; ARND=Alcohol-Related Neurodevelopmental Disorder; FAS = Fetal Alcohol Syndrome; TLD = Typical language development; HES = hesitation; INT = interjection; REV = revision; UW = unfinished word; WR = word repetition; SeR = segment repetition; SoR = sound repetition; PAU = pause; WM = words per minute; SM = syllables per minute

Table 2. Median and dispersion of values regarding disfluencies and speech rate in story oral narrative of individuals with Fetal Alcohol Spectrum Disorder and with typical language development

Typology of disfluencies	ARND		TLD		p-value
	Median	Min-Max	Median	Min-Max	
Typical disfluencies					
HES	2.8	1.4-4.7	1.0	0.0-2.0	0.002*
INT	0.0	0.0-3.1	0.0	0.0-1.6	1.000
REV	1.2	0.3-2.0	1.0	0.4-1.6	0.401
UW	0.1	0.0-0.7	0.4	0.0-0.9	0.145
WR	0.2	0.0-1.7	0.2	0.0-1.9	1.000
SeR	0.0	0.0-0.2	0.0	0.0-0.4	0.426
% typical disfluency	5.8	2.0-7.0	2.8	1.1-5.5	0.013*
Stuttering disfluencies					
SoR	0.0	0.0-0.1	0.0	0.0-0.0	1.000
PAU	2.8	0.8-5.6	0.4	0.0-1.8	0.003*
% stuttering disfluency	2.8	1.0-5.7	0.4	0.0-2.6	0.005*
Speech rate					
WM	55	40.3-118.2	83.4	65.6-140	0.063
SM	102	70-216	149	103-280	0.061

* Significant values (p≤0.05) – Mann-Whitney Test

Note: ARND= Alcohol-Related Neurodevelopmental Disorder; TLD = typical language development; Min = minimum; Max = maximum; HES = hesitation; INT = interjection; REV = revision; UW = unfinished word; WR = word repetition; SeR = segment repetition; SoR = sound repetition; PAU = pause; WM = words per minute; SM = syllables per minute

case S6 (ARND) when compared to S9 (FAS). On the other hand, the percentage of pause was higher for both cases with ARND (S5 and S6) when compared with cases with FAS (S8 and S9).

Even though pauses were among the stuttering disfluencies, according to the classification system adopted in this study, it was possible to note that their occurrence in narration has proven to have a similar function as hesitation, that is, to offer additional time to formulate what one wishes to say. Thus, we understand that its occurrence, not associated with typical stuttering disfluencies, may be more related to the difficulty in spoken language process in the level of conception and lexical selection.

The increase in such disfluencies in oral narration has been studied along the years, assuming that these ruptures associated to the narrative are important signs of the cognitive mechanisms involved in the spoken language production process[12,13]. In this perspective, both silent pauses and filled pauses (e.g. hesitation) have been adopted as signs of need of additional time for planning verbal information, with silent pauses more related to utterance formulation problems[12-14], and hesitations to word finding problems (e.g. word choice) with the wording already in progress[12]. Therefore, individuals with FASD may be said to present more difficulty in the construction of fluent utterances compared with their counterparts with TLD.

The occurrence of hesitation and silent pause in narration by the group with FAS suggested that these individuals present more difficulty in establishing a neuro-linguistic system for spoken language fluency - which often takes place in the first years of life[14] – which may be justified by the presence of impairments that exceed the difficulties in the executive functions mentioned in the literature.

Research on verbal fluency of individuals with FASD suggests that one common characteristic of these individuals is the presence of low rates in semantic and phonological evocation tests, regardless of whether they have the FAS diagnosis or not[8,9] that is, verbal fluency difficulties configure a common manifestation of the FASD phenotype as part of the difficulties in executive functions. These studies offer important information regarding the executive function mediating the spoken language production process, more specifically related to semantic evocation, an ability shown to be rather impaired in individuals with FASD. Thus, access to semantic representations is expected to be slower for individuals with FASD, which may also reflect in the processing speed of information required for the story generation, which require more complex cognitive and linguistic processes.

From the obtained data, it is possible to state that the information processing speed of these cases with FASD is impaired, since the number of syllables and words per minute was considerably lower compared with individuals with TLD. This data may be a reflection of a higher percentage of disfluencies for cases with FASD, contributing to a reduction of information flow produced in the narrative.

Even though this study did not focus on a more detailed analysis of the spoken language components – which may

be carried out in a future study for a more robust discussion regarding fluency aspects - it was possible to observe that, through a narrative task, cases with history of exposure to alcohol had more difficulties than their counterparts, both in the organization of the story narrative framework and the use of more complex syntactical and semantic structures. Therefore, the high occurrence of pause and hesitation is believed to be a result of the difficulties in spoken language, reflecting syntactical and lexical immaturity of these individuals, suggested in previous studies[5,8], as a result of impairment caused by alcohol consumption during fetal development, and also deriving from executive deficits,[8] since both dominions are important for narration.

CONCLUSION

The results of this study suggest that individuals with FASD present more hesitation-type disfluencies and silent pause in narration, when compared with the group with TLD, paired up for gender and chronological age.

Several studies have used the analysis of speech disfluencies as one of the parameters to be considered in the psycholinguistic and cognitive perspective of spoken language production models, since these characteristics may indicate difficulties presented by individuals in language formulation process. In case of individuals with FASD, disfluencies seem to be more related to difficulties in the conceptual plan and lexical selection or to difficulties in executive functions.

More specific studies are required to investigate possible relations between the occurrence of these disfluencies and the linguistic and cognitive aspects of narration, aiming to better characterize the singularities of subphenotypes constituting the Fetal Alcohol Spectrum Disorder, including speech fluency parameters.

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