Original Article

Artigo Original

Oral narrative of individuals with Fetal Alcohol Spectrum Disorder

Narrativa oral de indivíduos com Transtorno do Espectro Alcoólico Fetal

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Keywords

Language
Narration
Speech, Language and Hearing Sciences
Language Development Disorders
Fetal Alcohol Spectrum Disorder

ABSTRACT

Purpose: To investigate and compare the oral narrative of individuals with FASD and individuals with typical language development (TLD), as well as to correlate the narrative performance with the score from *4-Digit Diagnostic Code*. **Methods:** Participants were 20 individuals with FASD, of both genders, with chronological age between 6 and 16 years, and 20 individuals with TLD, same gender and similar to the FASD group in age and socioeconomic status. The oral narrative was elicited using the book *Frog, where are you?* and the data were analyzed in terms of macrostructure, microstructure and global coherence level. Measures regarding the macrostructure included the presence of typical structural elements of storytelling, while the microstructural aspects included words (total and different words), communication units (C-Units), lexical diversity, and mean length of C-Units. **Results:** Low performance was found in the FASD group for all macrostructural aspects, with the exception of linguistic markers. Among the microstructural aspects, lexical diversity and incomplete C-Units were different between the FASD and TLD groups. The FASD group presented lower global coherence level compared to the TLD group. Negative correlations were found between macrostructural aspects, facial characteristics, and Central Nervous System impairment. **Conclusion:** Restricted use of typical structural elements of storytelling with lower levels of coherence and reduced vocabulary distinguished the FASD from the TDL group. Future studies may explore whether the association between narrative performance and the 4-Digit Diagnostic Code items present predictive values in the narrative performance of individuals with FASD.

Descritores

Linguagem
Narração
Fonoaudiologia
Transtornos do Desenvolvimento da Linguagem
Transtorno do Espectro Alcoólico Fetal

RESUMO

Objetivo: Investigar e comparar a narrativa oral de indivíduos com Transtorno do Espectro Alcoólico Fetal (TEAF) e de indivíduos com desenvolvimento típico de linguagem (DTL) e correlacionar o desempenho na narrativa oral com a pontuação do "*4-Digit Diagnostic Code*". **Método**: Participaram 20 indivíduos com TEAF, de ambos os gêneros, com idade cronológica entre seis e 16 anos e 20 indivíduos com DTL, semelhantes quanto ao gênero, idade e nível socioeconômico aos do grupo TEAF. A narrativa oral foi eliciada por meio do livro "*Frog, where are you*?" e analisada quanto aos aspectos macroestruturais, microestruturais e nível de coerência global. Os aspectos macroestruturais incluíram elementos típicos de história e os microestruturais incluíram palavras (total, palavras diferentes), unidades comunicativas (C-Units), diversidade lexical e extensão média dos C-Units. **Resultados:** Desempenho inferior foi encontrado para o grupo TEAF em todos os aspectos macroestruturais, exceto para os marcadores linguísticos. Dentre os aspectos microestruturais, a diversidade lexical e a ocorrência de "*C-Units*" incompletos foram aspectos que diferenciaram os grupos TEAF e DTL. O grupo TEAF apresentou nível de coerência global inferior ao grupo DTL. Correlações negativas foram encontradas entre os aspectos macroestruturais e os itens características faciais e alterações no Sistema Nervoso Central. **Conclusão:** O uso restrito de elementos estruturais típicos de história com níveis inferiores de coerência e vocabulário reduzido diferenciaram o TEAF do DTL. Estudos futuros poderão explorar se a associação entre o desempenho narrativo e os itens do "*4-Digit Diagnostic Code*" apresentam valor preditivo no desempenho narrativo dos indivíduos com TEAF.

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Received: January 24, 2017

Accepted: March 15, 2017

Study carried out at Laboratório de Estudos, Avaliação e Diagnóstico Fonoaudiológico – LEAD, linked to the Post-Graduation Program in Speech-Language Therapy, Universidade Estadual Paulista – UNESP - Marília (SP), Brazil.

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Financial support: Fundação de Amparo à Pesquisa do Estado de São Paulo – FAPESP (process nº 2011/16672-2). This research was part of the scientific program of Instituto Nacional de Ciência e Tecnologia sobre Comportamento, Cognição e Ensino, with support from Conselho Nacional de Desenvolvimento Científico e Tecnológico (CNPq, Grant nº 465686/2014-1) and the Fundação de Amparo à Pesquisa do Estado de São Paulo (FAPESP, Grant nº 2014/50909-8).

Conflict of interests: nothing to declare.

INTRODUCTION

Fetal Alcohol Spectrum Disorder - FASD refers to a set of conditions resulting from pre-natal exposure to alcohol due to maternal consumption of the substance during pregnancy[1]. The term FASD encompasses different clinical subgroups depending on the condition and the degree of compromise, while Fetal Alcohol Syndrome - FAS is the most compromised subgroup with this clinical condition[2,3], characterized according to the diagnostic triad: (1) deficits in growth, (2) facial characteristics, and (3) Central Nervous System (CNS) impairments invariably associated with gestational exposure to alcohol.

Damage caused by alcohol varyingly compromise different aspects of neurodevelopment. In individuals with FASD, there have been frequent reports of deficits in problem-solving abilities, abstraction and elaboration of concepts[4], executive functions - particularly visuospatial[5] -, verbal and non-verbal working memory deficits[6,7], variable socio-behavioral impairment[8,9], and significant impairment in social and language skills that tends to be persistent thoughout life[10].

The language impairments described in the literature by studies with individuals with FASD are quite heterogeneous, both in terms of type and degree of compromise[11]. Such compromise is frequently characterized by significant deficits in language comprehension[12-15]. The studies investigating language production of individuals with FASD reported impaired phonological component[15], reduced expressive vocabulary for the age[12,16-19], production of vague, irrelevant, and inaccurate utterances[17]; difficulties to elaborate concepts and production of ambiguous statements [13,14], as well as increased frequency of hesitancy and silent pauses, which indicate significant difficulties to organize the story narrative scheme and use appropriate syntactic and semantic structures throughout the narrative[19].

Oral language impairments, particularly syntactic and semantic components, have been described as the most affected ones in FASD[2]. These changes were described in the studies focusing on assessment of spoken language by means of standardized language tests[16], as well as in those which prioritized naturalistic samples of spoken language, such as story narration[13,14,17,19].

Studies on the oral narrative of populations with FASD have been conducted and have shown the relevance of narrative performance for prognostic discussions, as well as for the identification of individuals with FASD at school age from specific measures for analysis of narrative coherence and cohesion[13]. Among the microstructural aspects of narrative, nominal agreement and referential cohesion mistakes are the most recurrent narrative difficulties of individuals with FASD, which help to distinguish narrative performance in their peers with typical development[14,20]. In addition, based on a study conducted, it seems that FASD phenotype manifestations present a significant association with the referential cohesion mistakes observed in these individuals' oral narrative. Upon data analysis, the increase in narrative cohesive problems presented an association with the severity of the specific diagnostic items investigated (presence of facial characteristics and CNS impairment) by the 4-Digit Diagnostic Code[20].

As shown, it is noted that, although some studies reported clinical manifestations that signal the difficulties presented by individuals with FASD in oral narrative, as to the microstructural aspects of narrative[13,14,17] and fluency of spoken language[19], little is known about the aspects that refer to the macrostructural dimension and global coherence of narrative. Also, although the association between the microstructural aspects (referential cohesion) of narrative and the items of the 4-Digit Diagnostic Code (facial characteristics and CNS impairment) has been reported as part of the FASD phenotype[20], such findings are still to be further explored.

Thus, the objective of this study was to investigate and compare the oral narrative of individuals with Fetal Alcoholic Spectrum Disorder (FASD) to individuals with typical language development (TLD) and correlate oral narrative performance with the score of the 4-Digit Diagnostic Code (growth deficit, facial characteristics, CNS impairment, and alcohol consumption during pregnancy).

METHODS

Individuals

20 individuals diagnosed with Fetal Alcoholic Spectrum Disorder (FASD) participated in this study and were compared to 20 individuals with Typical Language Development (TLD). The individuals were similar in gender, chronological age, and socioeconomic level. Each group (FASD and TLD) was composed of 12 female individuals (60%) and 8 male individuals (40%) with chronological age between 6 and 16 years (M=10.7; SD= 3.33). The socioeconomic classification of participants was similar, varying between classes A2 and D.

In order to perform the diagnostic classification of participants with FASD, a clinical geneticist applied the 4-Digit Diagnostic Code[21]. This is a tool developed by University of Washington FAS Diagnostic and Prevention Network to help diagnose FASD conditions. The instrument provides greater accuracy of clinical diagnosis based on a quantitative scale of risk signals structured according to the diagnostic triad and adding the confirmation of maternal alcohol use, that is: (1) growth deficits; (2) facial characteristics; (3) CNS impairment; and (4) alcohol consumption during pregnancy.

The inclusion criteria determined for the participants in the FASD group were: (1) having the diagnosis of FASD confirmed by a clinical geneticist; (2) not presenting a history of drug use simultaneously with alcohol consumption during pregnancy; (3) presenting a diagnostic classification based on the 4-Digit Diagnostic Code[21]; (4) presenting auditory thresholds within the normal parameters, below 25 dBNA; (5) presenting intelligible speech that ensured the evaluator's access to the content of oral narrative (words or utterances); (6) presenting a sample of oral narrative in accordance with the minimum structural criteria of a story narrative that could be assessed as to macrostructural aspects predicted in this study.

The presentation of a setting (reference to space, time, and an introduction of characters) and one or more episodes was adopted as the concept of the story-type narrative. An episode

is formed by three basic elements: a problem, the attempts to solve the problem, and a consequence of the problem[22]. Presence of conventional markers signalizing the beginning of a story (e.g. Once upon a time...) and its ending (e.g. ...and they lived happily ever after.); thus, a beginning, middle, and end[23].

The inclusion criteria for the TLD group were the absence of: (1) clinical signs suggesting syndromic condition of genetic or environmental etiology (e.g. rubella, toxoplasmosis, alcohol, drugs); (2) history of psychiatric or neurological conditions; (3) impairment in neuromotor, cognitive, and language development; and (4) presenting a sample of oral narrative in accordance with the minimum criteria of story narration.

Procedures

This study was approved by the Research Ethics Committee-CEP (Process number 0442/2012), and participation of individuals was authorized by parents and/or legal guardians upon signing the Informed Consent.

In order to elicit the oral narrative, the story book *Frog, where are you?*[24] was used. This book is composed of 24 scenes in black and white, without written words. All of the narrations were recorded by means of software *Praat (version 5.2.01)*. All of the narratives were fully transcribed for further analysis of macro and microstructural aspects adopted in this study.

The measures referring to the macrostructural aspects of the narrative included the presence of typical structural elements and linguistic markers in the story scheme, according to *Gramática de Histórias* [Grammar of Stories][22]. The narratives were punctuated and analyzed based on the presence of information referring to five structural elements: (1) setting, (2) theme, (3) plot, (4) misadventure, and (5) resolution, as proposed by Rossi et al.[25], in order to determine a partial score, for each element analyzed, and a global score. The narrative's global score was determined from the addition of partial scores, where the maximum score allowed was 31.

The global coherence analysis of the narrative was conducted based on the proposal by Spinillo and Martins[26], which consider as analysis parameters the maintenance of main characters and the theme/topic thoughout the narrative, the main event/plot, or situation-problem, and outcome. From this information, narratives were classified at four increasing levels of complexity that indicate the global coherence levels of the story.

The microstructural aspects analyzed were defined based on the proposals of Justice et al.[27] and Miller and Iglesias[28], which include: measure of linguistic productivity of the narrative, referring to the total number of words, total number of different words, and the number of communicative units of words; and measures of linguistic complexity of the narrative, which include lexical diversity, mean length of communication units in words, and number of complex communication units.

In order to analyze the linguistic productivity and complexity in terms of utterances, this study adopted segmentation of the narrative into communicative units named *"Communication Units - C-units"*. Each Communication Unit corresponds to a main clause (syntactically independent) and all of its dependent clauses present throughout the entire sample[28].

The data obtained were statistically analyzed. For comparison of continuous variables non compliant with normality, the non-parametric test Mann-Whitney was used to compare two independent samples (FASD and TLD). For analysis of correlation between two variables 4 digits, the Spearman correlation coefficient was used for ordinal variables. This study adopted a level of significance of 5% (0.050) both for the comparison and correlation analyses.

RESULTS

The results of the oral narrative comparative analysis of groups FASD and TLD indicated statistically significant differences between the groups, both for the macrostructural aspects (Table 1) and for the microstructural aspects of the oral narrative (Table 2). Table 1 shows that, among the typical story elements analyzed, only the linguistic markers that signal opening and closing of the story did not present a statistically significant difference between the groups. With regard to microstructural aspects, a statistically significant difference was only found for the item "lexical diversity" (Table 2).

Another aspect analyzed from the oral narratives presented by individuals in the FASD and TLD group was the occurrence of incomplete C-Units, which was higher in the FASD group compared to the TLD (Md_{GA}=4.0, Md_{GC}=0.5, p=0.003).

As to the classification of the oral narrative's global coherence level, it was verified that 45% of narratives in the FASD group were classified as Level II, and 30%, as Level I. The most complex levels were observed in only 20% (Level III) and 15% (Level IV) of the sample. On the other hand, half of the individuals in the TLD group were classified as Level III, and the other half, as Level IV.

Table 3 shows the results of the correlation analysis between the four items composing diagnosis of the 4-Digit Diagnostic Code (growth deficit, facial characteristic, impairment in CNS, and alcohol consumption during pregnancy) and the macro and microstructural aspects of oral narrative. It was possible to identify that the items "facial characteristics" and "CNS impairment" presented a statistically significant correlation, especially with regard to the macrostructural aspects of narrative.

It should be noted that the correlations found were all negative, indicating that the higher the level of impairment presented for the diagnostic item in question, the worse the performance presented by the individuals in the macrostructural aspects analyzed. Statistically significant correlations with the four characteristics composing the FASD diagnostic were not observed (Table 3).

DISCUSSION

This study proposed to investigate and compare the oral narrative of individuals with Fetal Alcohol Spectrum Disorder (FASD) and of individuals with typical language development (TLD). The results found indicated significant differences between the FASD and TLD groups in both the macro (Table 1) and microstructural aspects of oral narrative (Table 2).

Table 1. Comparison of FASD and TLD groups as to macrostructural aspects of oral narrative

Macrostructural aspects	Group	M	SD	Md	Min.	Max.	p
Typical story elements							
Setting	AG	3.45	0.69	3.00	2.00	5.00	0,017*
	CG	4.25	1.12	4.00	3.00	7.00	
Theme	AG	1.55	0.69	2.00	0.00	2.00	0.004**
	CG	2.00	0.00	2.00	2.00	2.00	
Plot	AG	1.95	1.70	2.00	0.00	6.00	<0.001**
	CG	4.95	1.50	5.00	2.00	8.00	
Misadventures	AG	2.80	1.61	3.00	0.00	6.00	<0.001**
	CG	5.95	1.67	6.00	3.00	8.00	
Resolution	AG	0.80	0.77	1.00	0.00	2.00	<0.001**
	CG	1.90	0.31	2.00	1.00	2.00	
Markers	AG	0.35	0.59	0.00	0.00	2.00	0.364
	CG	0.50	0.61	0.00	0.00	2.00	
Global score	AG	10.90	4.22	11.00	4.00	18.00	<0.001**
	CG	19.55	2.19	19.00	17.00	24.00	

Mann-Whitney Test; p<.05*; p<.005**
Caption: M = Mean; SD = Standard Deviation; Min. = Minimum; Max. = Maximum

Table 2. Comparison of FASD and TLD groups as to microstructural aspects of oral narrative

Microstructural aspects	Group	M	SD	Md	Min.	Max.	p
Linguistic productivity							
Total number of words	FASD	256.70	103.04	251.50	94.00	510.00	0.636
	TLD	239.50	74.27	233.00	138.00	401.00	
Total number of different words	FASD	92.70	28.45	86.00	46.00	162.00	0.213
	TLD	99.75	21.63	94.00	68.00	146.00	
Total number of C-Units	FASD	41.10	15.94	39.50	14.00	74.00	0.490
	TLD	38.70	11.49	36.50	21.00	62.00	
Linguistic complexity							
Lexical diversity	FASD	0.38	0.10	0.36	0.28	0.76	0.010*
	TLD	0.43	0.07	0.41	0.28	0.55	
Mean length of C-Units	FASD	6.37	1.07	6.64	4.85	9.50	0.298
	TLD	6.17	0.44	6.26	5.12	6.93	
Number of complex C-Units	FASD	3.65	3.42	3.50	0.00	10.00	0.191
	TLD	5.05	3.56	4.50	0.00	14.00	

Mann-Whitney Test; p<.05*; p<.005
Caption: M = Mean; SD = Standard Deviation; Min. = Minimum; Max. = Maximum

The differences found in the macrostructural aspects investigated were marked by the lower performance of the FASD group in partial scores attributed to the structural elements "setting", "theme", "plot", "misadventures", "resolution", global score, and global coherence level of the narrative (Table 1).

It is worth mentioning that the information regarding the setting is one of the first to be used by the child in narration, possibly because such information is presented in the first part of the narration to situate listeners in time, place, and with regard to the characters of the story. However, this item presented low scores even for adolescents with FASD who participated in this study. Thus, since individuals with FASD had difficulties to present information about the beginning of the story, it was also expected that difficulties would be found in the other parts of the narration, in which it was necessary to present not only the problem of the story, but also the characters' actions to resolve this problem, in a coherent logical and temporal sequence. Such impairments were evident not only by the low scores presented by individuals with FASD in the items "theme", "plot", "misadventures" and "resolution", but also in the global narrative analysis to classify the level of coherence.

In this study, both groups, FASD and TLD, presented restricted use of conventional linguistic markers, especially to introduce the stories. This finding justifies the similarity found between groups in this item of narrative analysis, as presented in the results (Table 1). A possible explanation for this finding, although applied only to the TLD group, may be due to the age of participants. Over the years, as individuals reach more organized and complex levels of story, these markers can be informally left aside without hampering the narrative's structure and coherence. However, the same speculation cannot be attributed to the FASD group, as the individuals presented difficulties to introduce and finish the oral narrative, as well as lower levels of coherence.

Table 3. Correlation between the typical story elements, global coherence of the narrative and characteristics of the 4-Digit Diagnostic Code presented by the FASD group

Typical story elements		Growth deficit	Facial characteristics	CNS impairment	Alcohol consumption
Setting	Coef. Correl. (r)	+0.125	-0.104	-0.023	-0.048
	Sig. (p)	0.601	0.664	0.924	0.840
Theme	Coef. Correl. (r)	-0.047	-0.370	-0.492	-0.129
	Sig. (p)	0.845	0.109	0.028*	0.589
Plot	Coef. Correl. (r)	-0.239	-0.458	-0.572	-0.067
	Sig. (p)	0.311	0.042*	0.008*	0.780
Misadventures	Coef. Correl. (r)	-0.034	-0.123	+0.083	+0.264
	Sig. (p)	0.888	0.605	0.728	0.260
Resolution	Coef. Correl. (r)	+0.164	-0.506	-0.357	0.000
	Sig. (p)	0.489	0.023*	0.123	≤0.999
Markers	Coef. Correl. (r)	+0.013	+0.075	-0.078	-0.270
	Sig. (p)	0.957	0.753	0.744	0.249
Global score	Coef. Correl. (r)	-0.032	-0.371	-0.349	-0.076
	Sig. (p)	0.895	0.107	0.131	0.749
Level of coherence					
Global coherence	Coef. Correl. (r)	-0.095	-0.445	-0.339	+0.127
	Sig. (p)	0.691	0.049*	0.143	0.593

Spearman correlation; p<.05*; p<.005
Caption: Coef. Correl. (r) = correlation coefficient; Sig. (P) = p-value

With respect to the level of coherence of the narrative, it was observed that the narration of individuals with FASD presented low thematic linearity and difficulty to sequentialize the events narrated, thus hampering the comprehension of the story by the evaluator. As shown, most of the individuals with FASD had their stories classified at the lowest levels of coherence (Level I, 30%, Level II, 45%). On the other hand, half of the individuals in the TLD group presented a narrative classified at Level III (50%), and the other half, at Level IV (50%).

The method of classification adopted in this study (see Spinillo and Martins[26]) assumes that coherence is to a large extent determined by the presence and organization of typical elements of story in organizing a theme. Thus, taking into consideration that lower narrative scores were found for individuals with FASD, they were also expected to present lower levels of story coherence compared to their peers.

As presented, this study also proposed to investigate the correlation between performance in oral narrative and the score in the items composing diagnosis of the 4-Digit Diagnostic Code (deficit in growth, facial features, CNS impairment, and alcohol consumption during pregnancy). The correlations were all negative, indicating that the higher the CNS impairment, or the more evident the facial dysmorphism, the lower the score obtained by the participant, both for narrative scheme and narrative coherence classification level. Correlations with statistical significance were found only for the structural elements "theme", "plot", and "resolution". These elements are considered central in the classification system of the coherence level proposed by Spinillo and Martins[26], which justifies the correlations found.

As to the results obtained from the comparison between individuals with FASD and with typical development (TLD) with regard to the microstructural aspects of oral story narrative, lower values were observed for the FASD group in relation to the TLD group in the items "total number of words", "total number of C-Units", and "mean length of C-Units". However, it was observed that the groups differed only in terms of lexical diversity of the narrative (Table 2), which is a linguistic measure that estimates the lexical proficiency or variety of different words spoken by the individual in communicative units[17].

By means of the data on lexical diversity, it was observed that the individuals with FASD presented lower performance in lexical proficiency, i.e. they used different words less often in relation to the total of words emitted when compared to the individuals with TLD. Reduced semantic repertoire has been described in previous studies on the language of children exposed to alcohol during pregnancy[12,16-19].

It was also observed that, with the increase in the number of utterances in the sample, words tended to be repeated, decreasing the rate of lexical diversity. This finding was observed in both the FASD and the TLD group, which corroborates a study carried out with children with typical language development[29].

Another aspect analyzed from the story narratives presented by the FASD and TLD individuals was the occurrence of incomplete "C-Units". The number of incomplete "C-Units" is not a formal measure of microstructural analysis, but a characteristic of the type of utterance produced throughout the narrative. It was observed that this characteristic was recurrent in the narration of individuals in both groups (FASD and TLD), although the number was significantly higher for the FASD. It is speculated that the greater occurrence of incomplete utterances in the narration of individuals with FASD may indicate the difficulty that individuals often present in planning and elaboration of the utterances, either in lexical selection or in semantic organization of the words that composed the ongoing utterance as of later

statements, which should be related with one another in order to maintain the meaning of the narrative.

Syntactic and semantic impairments have been reported in studies investigating the performance of individuals with FASD in language tests[12,15,16,18] or in more naturalistic language samples, such as oral narrative[11,13,14,19], since individuals with FASD needed revisions and reformulations of utterances throughout the oral narrative.

An important issue to be considered in this study and which may reflect on the findings regarding the microstructural aspects investigated is the material used to elicit the oral narrative. The picture book *Frog, where are you?*[24] favors productions with a logical and temporal sequence, resulting from the visual semantic repertoire offered by the pictures. Therefore, individuals both in the FASD and the TLD group presented more descriptive structures throughout narrations, with less complex syntactic structures, which ultimately disguises the microstructural aspects that may or not be impaired.

Another aspect investigated in this study was the existence of a possible relation between the macrostructural aspects of oral story narrative and the level of compromise of individuals with FASD, from the four key items composing the diagnostic criteria of the 4-Digit Diagnostic Code[21]. As mentioned, the higher the number of facial characteristics and signs of CNS impairment, the worse the performance in narrative of the more structural and organizational aspects of the story narrative scheme, which are found in the macrostructural dimension of the narrative. Our findings did not present association with microstructural aspects, unlike the findings of a previous study[20]. However, it is worth noting that the microstructural aspects contemplated in this study were different from the previous study[20], since this research did not contemplate an analysis of cohesive resources.

It is known that, in order to narrate a structured and coherent story, it is necessary to access a sort of mental representation of what a story narrative consists in and how it is structured. This narrative scheme, with its constituting elements, is gradually apprehended over the years, as the child is exposed to the story scheme model[23] and as the CNS matures[30]. The findings of functional resonance studies have shown that, both to understand and to produce narratives, it is necessary to activate different neural groups, which are not restricted to a single cerebral hemisphere[30]. Therefore, given the complexity of the task to orally narrate a story, it was expected that individuals with FASD participating in this study, with positive clinical findings more severe for CNS impairment, also presented more difficulty to perform the story narration.

CONCLUSION

The study has shown that individuals with FASD presented performance different from their peers with typical language development, characterized by restricted use of typical structural elements of the story narration scheme with lower levels of coherence and reduced vocabulary. It may be said that analysis of lexical diversity was the measure, among the ones contemplated in the microstructural aspects, that best discriminated individuals with FASD from their peers, which corroborated previous studies on the presence of significant deficits of semantic nature in this condition.

It is also concluded that the presence of incomplete utterances in the narration of individuals with FASD was an important and relevant characteristic to discuss semantic and syntactic deficits presented by these individuals. In addition, that such characteristic must be considered as part of the investigation items for oral narrative and intervention with further narrative difficulties.

Future studies may explore if the association found between narrative performance and the diagnostic items of the 4-Digit (facial characteristics and CNS impairment) present a predictive value for narrative development of individuals with FASD.

ACKNOWLEDGEMENTS

Fundação de Amparo a Pesquisa do Estado de São Paulo (FAPESP) for the support provided to carry out this research (process number 2011/16672- 2/MS/FAPESP), and Professors Drs. Antonio Richieri-Costa and Danilo Moretti-Ferreira for the genetic diagnosis of this study's casuistry.

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Author contributions

GG was responsible for the study design, data collection and analysis, and drafting of the paper; NFR and CMG were responsible for the study conception and design, data analysis and drafting of the paper.