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Original Article

Artigo Original

Fluency aspects of oral narrative task in del22q11.2 syndrome

Aspectos da fluência em tarefa de narrativa oral na síndrome del22q11.2

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Keywords

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ABSTRACT

Purpose: To investigate the fluency aspects of the oral narrative task in individuals with del22q11.2 syndrome and compare them with those of individuals with typical language development. **Methods:** Fifteen individuals diagnosed with del22q11.2 syndrome, both genders, aged 7-17 years participated in this study. They were compared with 15 individuals with typical language development, with similar gender and chronological age profiles. The oral narrative was elicited using the book "Frog, Where Are You?", and the fluency aspects were analyzed according to speech rate and type and frequency of disfluency (typical and stuttering). The number and duration of pauses were also investigated. The data were statistically analyzed. **Results:** The group with del22q11.2 syndrome showed a higher average when compared with the group without the syndrome for the percentage of typical disfluencies, mainly hesitation and revision. The group presenting the syndrome also showed a higher average for stuttering disfluencies, with pause as the most frequent disfluency. With respect to speech rate, the group with the syndrome presented a lower average for the number of words and syllables per minute. Individuals with del22q11.2 syndrome showed greater difficulties of narration than their peers. **Conclusion:** The fluency aspects of the oral narrative task in subjects with del22q11.2 syndrome were similar to those of individuals with typical language development regarding the presence of hesitation, revision, and pause, but they were different with respect to frequency of disfluency, which was higher in individuals with the syndrome.

Descritores

Narração
Linguagem
Fala
Medida da Produção da Fala
DiGeorge Syndrome

RESUMO

Objetivo: Investigar os aspectos da fluência em tarefa de narrativa oral na síndrome del22q11.2 e comparar com indivíduos com desenvolvimento típico de linguagem. **Método:** Participaram deste estudo 15 indivíduos com diagnóstico da síndrome del22q11.2, de ambos os gêneros, com idade cronológica de sete a 17 anos, que foram comparados a 15 indivíduos com desenvolvimento típico de linguagem, semelhantes quanto ao gênero e à idade cronológica. A narrativa oral foi eliciada com o livro *Frog Where Are You?,* e os aspectos da fluência foram analisados quanto ao tipo e frequência de disfluência (comum e gaga) e velocidade de fala. Foram analisados também o número e a duração das pausas. Os achados foram analisados estatisticamente. **Resultados:** O grupo com a síndrome del22q11.2 apresentou média superior em relação ao grupo sem a síndrome, para a porcentagem de disfluências comuns, principalmente hesitação e revisão. O grupo com a síndrome também apresentou média superior para disfluências gagas, sendo a pausa a disfluência mais frequente. Quanto à velocidade de fala, o grupo com a síndrome apresentou média inferior para o número de palavras e sílabas por minuto. Assim sendo, conclui-se que os indivíduos com a síndrome del22q11.2 apresentaram mais dificuldades para narrar do que os seus pares. **Conclusão:** Os aspectos da fluência investigados foram semelhantes entre os grupo com a síndrome del22q11.2 e com desenvolvimento típico de linguagem quanto à presença de hesitação, revisão e pausa na narrativa oral, porém distinto quanto à frequência dessas disfluências, que foi superior para os indivíduos com a síndrome.

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INTRODUCTION

The del22q11.2 syndrome is a neurodevelopmental disorder of genetic nature caused by microdeletion in the long (q) arm of chromosome 22 at the q11.2 band, with deletion of a region encompassing approximately 40 genes[1]. However, the clinical heterogeneity of this condition seems to justify the different denominations found in the current literature: (a) Sedlácková syndrome[2]; (b) DiGeorge sequence[3]; (c) velo-cardio-facial syndrome[4]; (d) CATCH22[5]; (e) del22q11.2 syndrome[6].

The del22q11.2 syndrome has a population frequency of approximately 1:2000 live births. This might be an underestimated prevalence because many cases are underdiagnosed due to mild phenotypic expression, which is clinically difficult to recognize. The most common phenotypic characteristics of this syndrome are cardiac abnormalities, hyperleptoprosopy, hooded eyelids, tubular nose with broad tip, and cleft lip and palate, including submucous cleft palate, retrognathism, and malocclusion[7].

Other manifestations related to the phenotype of this condition are speech, cognitive, learning, logical-mathematical reasoning, and behavioral disorders[8,9], including typical psychiatric manifestations such as attention deficit with hyperactivity disorder (ADHD), autism spectrum disorder (ASD), anxiety and mood disorders, psychotic disorders and schizophrenia[10], and significant loss of social cognition[11].

The neurocognitive phenotype of the del22q11.2 syndrome has also been studied by many researchers, because this syndrome is a neurogenetic condition in which individuals are likely to show a superiority of verbal intelligence over non-verbal intelligence, mainly of visuoconstructional nature; however, these individuals often present demoted performance in both tasks, verbal and non-verbal, compared with that expected for the chronological age[12].

Speech and language impairment has been described since the first years of life of these individuals[13]; however, these findings are divergent with respect to the performance of receptive and expressive language skills. Although receptive and expressive skills have been described as equally affected in individuals with this syndrome[14], studies have shown more significant impairment in receptive skills compared with expressive skills[15]. Also, studies have highlighted that deficit in the production of spoken language presented by individuals with del22q11.2 syndrome seem to exceed the anatomic changes of the phonoarticulatory organs[12,14,16]. Difficulties in more complex tasks, such as production of oral narrative[16] and verbal fluency, were described as part of the phenotype of this condition[17,18].

No studies have been found on the analysis of frequency and type of disfluency (typical and stuttering) and speech rate in oral narrative tasks in del22q11.2 syndrome in the national and international literature. In contrast, such aspects of speech fluency in genetic syndromes, particularly in the oral narrative situation, have been an important resource for the characterization of changes in spoken language in genetic conditions such as Williams-Beuren syndrome[19-21] and Fetal Alcohol Spectrum Disorder[22].

This study aimed to investigate the fluency aspects of the oral narrative task in individuals with del22q11.2 syndrome and compare them with those of individuals with typical language development.

METHODS

Sample

Participants signed an Informed Consent Form (ICF) developed according to the Resolution MS/CNS/ CNEP n. 196/96 of 10 October 1996, and approved by the local Research Ethics Committee (IRB) (n. 0142/2011).

The study sample included 30 individuals who were divided into two groups: Study Group (SG), comprising 15 subjects aged 7-17 years (mean = 11.6; SD = 2.6) with positive results in the Fluorescent *In Situ* Hybridization (FISH) test for deletion of chromosome 22 in the q11.2 band; and Control Group (CG), composed of 15 individuals aged 7-17 years (mean = 11.7; SD = 2.7) with typical language development. The groups were paired for gender and age range for comparison of the findings.

The selection of individuals with del22q11.2 syndrome (SG) was conducted at the Hospital for Rehabilitation of Craniofacial Anomalies, University of Sao Paulo - USP according to the following inclusion criteria: (a) positive result in the FISH test for deletion of chromosome 22 in the q11.2 band; (b) chronological age between six and 18 years.

The CG group included individuals selected in municipal and state schools located in the municipality of Marilia, Sao Paulo state. Selection was authorized by the Department of Education and was conducted according to the following established criteria: (a) absence of difficulty in spoken language; (b) absence of low school performance; (c) gender and chronological age pairing with the SG individuals; (d) absence of speech therapy or educational referrals and/or treatments.

All participants presented audiometric thresholds below 25 dBNA[23]. SG individuals had Intelligence Quotient (IQ) total scores between 68 and 110, whereas CG individuals had IQ total scores between 90 and 110. The Wechsler Intelligence Scales for children (WISC-III)[24] and for adults (WAIS-III)[25] were used to establish the IQ total scores.

Procedure for characterization and analysis of fluency profile in oral narrative

The book "Frog, where are you?"[26] was used to elicit the oral narrative. This book is composed of 29 illustrations without text displayed in sequence. The individuals were requested to hold the book and were instructed to flip through every page of it and then tell the history; after that, they were instructed to tell the best history possible using the book. All samples were recorded (audio and video) using a SONY - DCR-SX83 camcorder and a TSI - MS115 MCL-VHF wireless headset attached to a portable digital recorder.

For analysis of the fluency aspects, the sample of oral narrative was transcribed according to the criteria established

in the Teste de Linguagem Infantil - ABFW, Fluency Area[27]. These criteria consider (a) type and frequency of disfluencies subdivided into typical (e.g., hesitations, revisions, and unfinished words) and stuttering (e.g., repetition of sounds and/or syllables (two or more), and/or words; blocks, and pauses); b) speech discontinuity by the sum of typical and stuttering disfluencies, or stuttering disfluencies (in percentage); (c) and speech rate, which analyzes the number of words and syllables per minute. This study considered a fourth item as part of the fluency analysis: the percentage of typical disfluencies, calculated from the total of typical disfluencies.

The software PRAAT, version 4.3.27 was used for the recording and analysis of the following aspects: (a) total duration of oral narrative, (b) number of pauses, and (c) duration of pauses in the narrative.

Ruptures in the oral narrative lasting ≥ 2 seconds (≥ 0.03 minutes) were considered as pauses according to the ABFW test standards[27].

The quantitative variables were tested for normality of data distribution using the Shapiro-Wilk test. The parametric Student's t-test was used for normal distribution data for two independent samples. The nonparametric Mann-Whitney test was used for the data which did not comply with normality for two independent samples.

RESULTS

Table 1 presents the descriptive statistical values for typical disfluency, specifying the percentage found for each disfluency type for the SG and CG. SG individuals showed higher mean value for all values of typical disfluencies compared with those of the CG, except for repetition of phrases, which was not found in either group. It is worth noting that hesitation and revision were typical disfluencies that appeared more frequently in the oral narrative in both the SG and CG, but they occurred more frequently in the SG than in the CG.

Table 2 shows the descriptive statistical values for stuttering disfluency percentage in the SG and CG. In the CG, only one subject presented stuttering disfluency of the pause type.

Table 1. Percentage of typical disfluencies found in the oral narrative of the Study Group (SG) and Control Group (CG)

Disfluencies Typical	Group	Min.	Mean	Median	Max.	SD	CV
Hesitation	SG	0.00	2.78	2.22	7.35	2.03	73.00
	CG	0.00	1.17	0.86	5.51	1.41	120.37
Interjection	SG	0.00	0.19	0.00	1.00	0.35	187.23
	CG	0.00	0.04	0.00	0.47	0.13	295.46
Revision	SG	0.00	0.91	0.95	1.93	0.64	70.34
	CG	0.09	0.79	0.81	1.77	0.44	55.96
Unfinished word	SG	0.00	0.44	0.14	3.03	0.80	178.95
	CG	0.00	0.25	0.21	0.86	0.26	102.48
Word repetition	SG	0.00	0.38	0.32	1.31	0.40	105.66
	CG	0.00	0.27	0.00	1.99	0.53	197.88
Segment repetition	SG	0.00	0.06	0.00	0.30	0.11	180.49
	CG	0.00	0.00	0.00	0.00	0.00	N.C.
Phrase repetition	SG	0.00	0.00	0.00	0.00	0.00	N.C.
	CG	0.00	0.00	0.00	0.00	0.00	N.C.

Caption: Min. = Minimum; Max. = Maximum; SD = Standard deviation; CV = Coefficient of variation (percentage); N.C. = Not Calculated

Table 2. Percentage of stuttering disfluencies found in oral narrative of the Study Group (SG) and Control Group (CG)

Disfluencies Stuttering	Group	Min.	Mean	Median	Max.	SD	CV
Syllable repetition	SG	0.00	0.03	0.00	0.49	0.13	387.3
	CG	0.00	0.00	0.00	0.00	0.00	N.C.
Sound repetition	SG	0.00	0.00	0.00	0.00	0.00	N.C.
	CG	0.00	0.00	0.00	0.00	0.00	N.C.
Prolongation	SG	0.00	0.05	0.00	0.63	0.16	313.19
	CG	0.00	0.00	0.00	0.00	0.00	N.C.
Block	SG	0.00	0.00	0.00	0.00	0.00	N.C.
	CG	0.00	0.00	0.00	0.00	0.00	N.C.
Pause	SG	0.00	0.35	0.00	2.27	0.68	194.68
	CG	0.00	0.004	0.00	0.06	0.02	387.30
Segment insertion	SG	0.00	0.00	0.00	0.00	0.00	N.C.
	CG	0.00	0.00	0.00	0.00	0.00	N.C.

Caption: Min. = Minimum; Max. = Maximum; SD = Standard deviation; CV = Coefficient of variation (percentage); N.C. = Not Calculated

SG and CG individuals were compared with respect to the total percentage of typical, stuttering, and speech discontinuity disfluencies using the Mann-Whitney test (Table 3). The results showed that both groups presented statistically significant differences. In Table 3, it is possible to observe that the percentage of disfluencies (typical, stuttering, and speech discontinuity) was higher for individuals with del22q11.2 syndrome compared with that for individuals with typical language development, with statistically significant difference (Table 3).

Regarding speech rate (words per minute and syllables per minute), the statistical values found for the number of words per minute and syllables per minute identified a statistically significant difference in the SG compared with the CG (Table 4). Individuals with del22q11.2 syndrome (SG) showed a lower mean for number of words and syllables per minute than CG individuals.

DISCUSSION

Investigation and comparison of the aspects of fluency in oral narrative in individuals with del22q11.2 syndrome (SG) and with typical language development (GC) allowed for identification of similarities and differences between the groups.

The study group presented a profile similar to that of the control group with respect to the type of typical disfluencies that occurred in oral narrative (hesitation, interjection, revision, unfinished word, and repeated word), with hesitation and revision presenting a higher frequency in both groups. In contrast, differences between the groups were observed with respect to the frequency of occurrence of oral narrative disfluencies, with higher percentage in the SG compared with the CG, which explains the significant difference found between the groups; as well as to the total percentage of typical narrative disfluencies, which was higher in the SG.

Typical disfluencies such as hesitation and revision, also called fillers, are used by the speaker as a time resource for interpersonal coordination[28]. It is worth mentioning that hesitation and revision are commonly observed in the speech of all individuals, with or without a language problem, not determining therefore a fluency disorder[27].

Nevertheless, typical disfluencies such as hesitation, interjection, revision, and repetitions may represent uncertainties within spoken language production[29]. A higher frequency of these ruptures has been observed in the oral narrative of individuals with genetic neurodevelopmental disorders that occur with intellectual impairment[19-22], as in our sample, as well as in that of individuals with specific language disorders[30].

In addition to the frequency of the aforementioned typical disfluencies, another aspect that contributes to the difference between the fluency profiles of the groups investigated is the occurrence of stuttering disfluencies in narration, especially syllable repetition and prolongation, which were observed only in the SG. It is worth remarking that the percentages found for disfluency in the SG were less frequent than five occurrences (syllable repetition and prolongation), a value expected for individuals without fluency pathology[29].

In contrast, both groups (SG and CG) presented stuttering disfluency of the pause type, with higher percentage observed in the SG individuals. More than five occurrences were observed only for the pause disfluency in the oral narrative of both groups. Although pause is classified as a stuttering disfluency according to the taxonomy suggested by the ABFW instrument[27], adopted in this research, it is possible to infer that the silent pause can also be used to allow the individual more time to complete the message or introduce new information, similarly to hesitation[28].

Finally, another factor that contributed to differentiate the fluency profile of the groups was the speech rate. As shown in Table 4, the SG presented lower values compared with those of

Table 3. Comparison between the total percentage of typical, stuttering, and speech discontinuity disfluencies of the Study Group (SG) and Control Group (CG)

Percentage of Disfluency	Group	Min.	Mean	Median	Max.	SD	p
Typical	SG	0.96	4.77	4.85	8.82	2.44	0.008957*
	CG	0.20	2.52	1.89	7.40	2.05	
Stuttering	CG	0.00	0.43	0.16	2.27	0.66	0.004173**
	CG	0.00	0.004	0.00	0.06	0.02	
Speech discontinuity	CG	0.96	5.20	5.08	9.31	2.58	0.005442**
	CG	0.20	2.52	1.89	7.40	2.06	

Caption: Mann-Whitney test; Min. = Minimum; Max. = Maximum; SD = Standard deviation. *p ≤ 0.05. **p ≤ 0.005

Table 4. Sistical values of speech rate for the Study Group (SG) and Control Group (CG)

Speech rate	Group	Min.	Mean	Max.	SD	p
Words per minute	SG	40.68	70.67	140.90	27.04	0.003171**
	CG	72.45	94.26	125.80	12.93	
Syllables per minute	SG	46.31	110.80	154.20	33.84	0.0001341**
	CG	114.80	161.50	224.40	32.67	

Caption: Student's t-test; Min. = Minimum; Max. = Maximum; SD = Standard deviation. **p ≤ 0.005

the GC for number of words and number of syllables per minute. This result was expected because speech rate parameters are related to the number of words and, hence, of fluent syllables in the narrative, so that increased frequency of both silent pauses and hesitation and revision interferes with the counting of syllables and words per minute, resulting in a decreased speech rate, as seen for individuals with del22q11.2 syndrome in this study.

Similar features were reported in studies using the same methodology of this study, as seen in individuals with Williams syndrome[19-21] and Fetal Alcohol Spectrum Disorder[22]. The decreased speech rate and verbal information quality produced by individuals with the del22q11.2 syndrome compared with those of individuals with typical language development has been reported in studies using semantic recall tasks as measures of verbal fluency[17,18]. However, to our knowledge, this is the first study that analyzed fluency under the perspective of type and frequency of disfluencies, as well as of speech rate in the oral narrative.

CONCLUSION

The fluency aspects of the oral narrative task in subjects with del22q11.2 syndrome were similar to those of individuals with typical language development regarding the presence of hesitation, revision, and pause, but they were different with respect to frequency of disfluency, which was higher in individuals with the syndrome. This finding may explain the reduced speech rate in the oral narrative of individuals with the syndrome as for the number of syllables and words per minute.

The speech fluency data suggest that the ruptures found in the oral narrative may be more closely related to problems encountered in language conceptualization and formulation than to difficulties in articulation (phonological and motor).

The analysis of speech fluency in the narrative of individuals with del22q11.2 syndrome can contribute to a better understanding of the language phenotype of this genetic condition, and also as one of the specific measures of oral narrative performance in these individuals, which would contribute to the planning of speech-language pathology intervention.

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Author contributions

AOS was responsible for the project ellaboration, collection and analysis of data, and writing of the manuscript; NFR and CMG were responsible for the study design, data analysis, co-orientation and orientation of the study, and writing of the manuscript; MCFFT was responsible for the statistical analysis of data, discussion of results, and writing of the manuscript; ARC was responsible for the genetic diagnosis, sample selection, and writing of the manuscript.