



Evaluation of Heart Rate Variability in Children with Stutter

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ABSTRACT

Aim: The autonomic nervous system has a direct or indirect effect on motor speech and its development. The results of studies evaluating autonomic functions in stuttering individuals show that further research is needed in different age groups. In this study, the aim was to evaluate autonomic function by analysing heart rate variability (HRV) in children with stutter.

Materials and Methods: In this study, a total of 41 individuals (11 females, 30 males) between the ages of 6-17 years (mean age: 10.17±2.75), diagnosed with stuttering were evaluated. The control (healthy/normal) group comprised 41 individuals (12 female, 29 male) between the ages of 6-17 years (mean age: 10.78±2.78), who did not have any speech disorder complaints and no family history. The level of stuttering was designated by applying the Turkish version of the Stuttering Severity Instrument Fourth Edition to the diagnosed group. All cases were tested for HRV and analysed using the 24-hour Holter electrocardiography recording method. Correlations between stuttering severity and the HRV parameters of the stuttering group, and correlations of HRV parameters in both groups were examined.

Results: A positive significant correlation was found between secondary behaviours in the stuttering group and the standard deviation of the mean NN intervals in 5-minute recordings (SDANN) of the HRV test. Additionally, when the correlation of HRV parameters between the groups was examined, the SDANN parameter in the stuttering group was statistically significantly higher ($p<0.05$). The other parameters were not statistically significantly different between the groups.

Conclusion: In this study, when the HRV parameters of the stuttering children were compared with the non-stuttering children, no significant differences were found to prove autonomic nervous system dysregulation.

Keywords: Stuttering, autonomic nervous system, heart rate variability

Introduction

Stuttering is a speech disorder which affects the normal flow of speech through the repetition or prolongation of sounds, syllables, or words. It can also be accompanied by secondary behaviours.

The autonomic nervous system controls involuntary movements and organ functions in the body and it plays a role in motor speech and development. Furthermore, emotions can cause physiological changes which activate the sympathetic part of the autonomic nervous system,

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which can disrupt speech patterns and affect the speed, rhythm, and fluency of speech (1,2).

Several theories have been proposed to explain the aetiology of stuttering. However, studies examining autonomic responses in individuals with stuttering problems are insufficient. Some studies have shown that a decrease in heart rate (HR) during speech tasks in adults with stuttering indicates that sympathetic arousal is accompanied by impairments in motor processes characteristic of stuttering (3,4). In their study, Doruk et al. (2) investigated the presence of autonomic nervous system dysregulation in young adult stutterers by analysing the results of 24-hour Holter electrocardiogram HR variability (HRV). Their study concluded that stuttering patients tended to have parasympathetic system dominance (2). In 2010, a study was conducted with young adult stuttering patients which obtained results supporting autonomic nervous system dysregulation through the application of the tilt test (5). In 2009, Tarkowski and Paprzcki (6) conducted a study which was not entirely compatible with previous research. They stated that autonomic responses were not significantly related to the fluency of speech (6).

Conture et al. (7) investigated childhood stuttering in order to determine whether emotional intelligence interacts with emotional stress. They found that increased sympathetic activity had a negative impact on speech (7). In 2016, Choi et al. (8) studied the relationship between mood, emotional stress, and stuttering in preschool children. They concluded that the autonomic nervous system plays a role in stuttering (8). Contrary to these findings, Walsh et al. (9) reported that speech-related sympathetic arousal was not a significant factor in early childhood stuttering. They suggested that developmental profiles in different age groups should be explored in future studies involving children who continue to stutter.

This study aimed to evaluate the severity of stuttering in school-age children and compare HRV between stuttering and healthy individuals. The hypothesis was that there are differences in HRV values between stuttering and non-stuttering children.

Materials and Methods

Study Design

This study was conducted jointly by the otorhinolaryngology and pediatric cardiology departments in accordance with the Declaration of Helsinki. The study was conducted with 41 patients who presented with stuttering complaints and met the inclusion criteria, as well

as 41 healthy individuals without stuttering complaints. All of the individuals were between the ages of 6-17 years. The Clinical Research Ethics Committee of Ege University Faculty of Medicine approved this study (approval no.: 16-9/1, date: 14.10.2016).

This study excluded those individuals with hearing impairments, those outside the 6-18 year age range, those with a history of cardiac and circulatory disease, head trauma, metabolic disease, convulsions or central nervous system infection, and those using psychiatric drugs.

The volunteers were divided into two groups: the control group, consisting of patients with normal hearing and no stuttering complaints, and the subject group, consisting of patients diagnosed with stuttering. All patients received an Informed Consent Form, Case Report Form, and a hearing assessment with pure tone threshold audiometry. The severity of stuttering was evaluated using the Turkish version of the Stuttering Severity Instrument (SSI-4TR), and HRV analysis was conducted with a 24-hour Holter application. The Holter was applied in the cardiology department on the first day, and the findings were recorded and extracted the following day (10).

Stuttering Severity Instrument Fourth Edition

Stuttering Severity Instrument Fourth Edition (SSI-4) is a scale widely used to assess the severity of stuttering. SSI-4TR was adapted by Mutlu et al. (11) in 2014. This tool can be used for both clinical and screening purposes. The severity of stuttering in children and adults is measured by evaluating four areas of speech behaviours: frequency, duration, secondary behaviours, and the naturalness of speech. Frequency is expressed as a percentage and converted into scale scores ranging from 2 to 18, duration is scored on a scale of 2 to 18, and secondary behaviours are scored on a scale of 0 to 20. The total score determines the severity of stuttering, with higher scores indicating greater severity. Naturalness of speech is rated on a visual analogue scale from 1 (very natural speech) to 9 (very unnatural speech) (10,11).

HRV

HRV reflects physiological changes in HR resulting from the interaction between the sympathetic and parasympathetic nervous systems. HRV records are obtained through 24-hour Holter electrocardiography. The records are first transferred to a computer environment and evaluated using the Holter program. They are then visually examined by an experienced professional, who excludes any parasitic areas from the evaluation.

On the first day of testing, a Holter device is installed in the cardiology department. The findings are recorded and extracted the following day and evaluated by experienced paediatric cardiology physicians.

Statistical Analysis

This study analysed data using the SPSS 20.0 program (IBM SPSS Statistics for Windows, Version 27.0. Armonk, NY: IBM Corp, USA). Gender and age distributions are presented through cross-tabs. Normal distribution was assessed using the Shapiro-Wilk test. For data without normal distribution, the Mann-Whitney U test was used for comparison. Continuous variables are presented as mean \pm standard deviation and categorical variables as numbers and percentages. Pearson's correlation analysis was used to examine relationships between continuous variables, while chi-square analysis was used to analyse differences between categorical variables. A p value <0.05 was considered statistically significant.

Results

This study analysed data from 41 patients diagnosed with stuttering and 41 healthy controls, a total of 82 individuals. Table I shows the demographic characteristics of the patients included in this study. The mean duration of stuttering at the time of diagnosis was 16 ± 4.7 months. There was no statistically significant difference between the two groups in terms of age or gender distribution. Analysis of family history in the stuttering group revealed that 16

individuals (39%) had a family history of stuttering, whereas 25 (61%) did not.

The severity of stuttering in the patient group ranged from mild in 1 patient (2.4%), moderate in 19 (46.3%), severe in 14 (34.1%), and to very severe in 7 (17.1%). The results of the assessment of stuttering frequency, duration, secondary behaviours and the naturalness of speech are given in Table II.

Upon examining the average distribution of the HRV parameters between the groups, the standard deviation of the mean NN intervals in 5-minute recordings (SDANN) values for the stuttering group were significantly higher than the control group ($p<0.05$). No significant differences were observed between the groups in terms of the other variables ($p>0.05$) (Table III).

When examining the correlation between reading, speaking, frequency, seconder behaviours, and duration values which make up the total SSI-4TR scores in the stuttering group, along with HRV parameters, a weak positive correlation was found between the secondary behaviour scores and the SDANN values, and a weak negative correlation was found between maxHR values and beats values ($p<0.05$) (Table IV).

When the correlation between the total score of SSI-4TR, severity and naturalness of speech values of the stuttering group were analysed with the HRV parameters, a weak negative correlation was found between the total score of SSI-4TR and beats values (Table V).

Table I. The demographic characteristics of the cases

	Stutter (n=41)	Control (n=41)	p value
Age (Mean \pm SD)	10.17 \pm 2.75	10.78 \pm 2.78	0.228
Gender n (%)			
Male	30 (73.2)	29 (70.7)	0.806
Female	11 (26.8)	12 (29.3)	0.317
SD: Standard deviation			

Table II. The mean of the parameters SSI-4 score in stuttering children

	Mean \pm SD	Median (Min.-Max.)
Frequency	15.24 \pm 1.9	15 (18-11)
Reading	7.49 \pm 1.19	8 (9-5)
Speaking	7.76 \pm 1.18	8 (9-4)
Secondary behaviours	6.21 \pm 3.82	6 (17-0)
Duration	7.29 \pm 2.4	8 (12-2)
Naturalness of speech	5.78 \pm 2.13	7 (9-2)
SD: Standard deviation, Max.: Maximum, Min.: Minimum, SSI-4: Stuttering Severity Instrument Fourth Edition		

Table III. The average distribution of HRV parameters according to the groups

	Control group	Stuttering group	Z	p value
	Median (Min.-Max.)	Median (Min.-Max.)		
MinHR	50 (40-70)	51 (40-67)	-0.851	0.395
AVGHR	83 (56-126)	85 (67-106)	-1.819	0.069
MaxHR	142 (106-256)	147 (120-178)	-1.412	0.158
Beats	113,080 (31,389-151,742)	116,727 (74,458-141,172)	-0.969	0.332
SDNN	152 (80-285)	152 (114-275)	-0.060	0.952
SDANN	123 (49-233)	132 (81-360)	-2.203	0.028
SDNNIndex	99 (41-210)	85 (52-163)	-1.271	0.204
RMSSD	112 (36-289)	93 (49-248)	-1.442	0.149
SDSD	112 (36-289)	93 (49-248)	-1.517	0.129
NN50	27.64 (4.04-55.2)	26.54 (7.65-65.95)	-0.107	0.915
pNN50	31.45 (11-58.71)	26.98 (14.32-55.51)	-0.645	0.519
TR.INDEX	34.88 (15.76-55.15)	38.6 (14.43-56.63)	-1.053	0.293
ULF	9,862.46 (2,558.84-30,595.06)	12,069.96 (3,222.52-36,116.41)	-1.526	0.127
VLF	2,637.94 (673.58-10,982.59)	2356.12 (894.44-5,194.71)	-0.858	0.391
LF	3,043.11 (400.29-21,651.75)	2234.54 (808.62-6,089.95)	-1.748	0.080
HF	3,596.94 (417.88-30,371.53)	2756.28 (1,240.19-9,719.89)	-1.118	0.264
TP	8,602.26 (1,254.81-58,280.7)	6430.95 (3,115.13-18,862.72)	-1.247	0.212
LF/HF	0.88 (0.5-1)	0.83 (0.34-1)	-1.925	0.054

Mann-Whitney U Analysis

Max.: Maximum, Min.: Minimum, MinHR: Minimum heart rate beats/min, MaxHR: Maximum heart rate beats/min, AVGHR: Mean heart rate beats/min, NN: Cycle length between two normal beats, SDNN: Standard deviation of all NN intervals during the examination, SDNN index: Mean of the standard deviations of all NN intervals in 5-min recordings, SDANN: Standard deviation of average NN intervals in 5-min recordings, NN50: Number of adjacent NN intervals with a difference of more than 50ms during the entire recording, pNN50: NN50 number divided by the total number of NNs, RMSSD: Square root of the sum of the differences of consecutive NN intervals in the 24-hour recording) TR.INDEX: Division of all NN interval number to the number of NN intervals in the mode length, HF: High-frequency band, LF: Low-frequency band, VLF: Very-low-frequency band, TP: Total power; variance of all NN intervals, ULF: Asleep, power in the low-frequency range

Table V. Correlation between stuttering children's SSI-4 Total score, severity and naturality of speech values with HRV parameters

	SSI-4 Total score		Severity		Naturality of speech	
	r	p value	r	p value	r	p value
MinHR	0.083	0.604	0.054	0.737	0.074	0.644
AVGHR	-0.062	0.699	-0.036	0.823	0.065	0.688
MaxHR	-0.273	0.084	-0.088	0.584	0.061	0.705
Beats	-0.317	0.044	-0.220	0.166	-0.101	0.529
SDNN	-0.013	0.935	0.006	0.970	-0.122	0.447
SDANN	0.269	0.089	0.263	0.096	0.187	0.241
SDNNIndex	0.048	0.766	0.025	0.877	-0.112	0.487
RMSSD	0.123	0.443	0.068	0.672	-0.077	0.634
SDSD	0.132	0.410	0.074	0.644	-0.071	0.658
NN50	-0.046	0.775	-0.020	0.901	-0.170	0.288
pNN50	-0.113	0.480	-0.090	0.576	-0.119	0.457

Table V. Continued

	SSI-4 Total score		Severity		Naturality of speech	
	r	p value	r	p value	r	p value
TR.INDEX	-0.041	0.797	-0.001	0.993	-0.113	0.483
ULF	-0.179	0.264	-0.157	0.328	-0.263	0.097
VLF	0.232	0.145	0.224	0.159	0.084	0.599
LF	0.219	0.169	0.175	0.273	0.099	0.539
HF	0.167	0.296	0.158	0.323	0.028	0.862
TP	0.209	0.189	0.188	0.240	0.062	0.701
LF/HF	-0.160	0.316	-0.207	0.194	-0.068	0.672

Pearson correlation
MinHR: Minimum heart rate beats/min, MaxHR: Maximum heart rate beats/min, AVGHR: Mean heart rate beats/min, NN: Cycle length between two normal beats, SDNN: Standard deviation of all NN intervals during the examination, SDNN index: Mean of the standard deviations of all NN intervals in 5-min recordings, SDANN: Standard deviation of average NN intervals in 5-min recordings, NN50: Number of adjacent NN intervals with a difference of more than 50ms during the entire recording, pNN50: NN50 number divided by the total number of NNs, RMSSD: Square root of the sum of the differences of consecutive NN intervals in the 24-hour recording, TR.INDEX: Division of all NN interval number to the number of NN intervals in the mode length, HF: High-frequency band, LF: Low-frequency band, VLF: Very-low-frequency band, TP: Total power; variance of all NN intervals, ULF: Asleep, power in the low-frequency range

Table IV. Correlation of stuttering children's reading, speaking, frequency, secondary behaviour, and duration values with HRV parameters

	Reading		Speaking		Frequency		Secunder behaviour		Duration	
	r	p value	r	p value	r	p value	r	p value	r	p value
MinHR	-0.307	0.051	0.221	0.165	-0.054	0.735	0.126	0.432	0.019	0.905
AVGHR	-0.104	0.519	0.304	0.053	0.124	0.439	-0.207	0.194	-0.032	0.842
MaxHR	-0.001	0.996	0.139	0.386	0.086	0.593	-0.393	0.011	-0.158	0.325
Beats	0.008	0.960	0.132	0.410	0.087	0.587	-0.455	0.003	-0.228	0.151
SDNN	0.002	0.990	-0.103	0.520	-0.063	0.695	-0.008	0.958	0.049	0.760
SDANN	-0.190	0.233	0.064	0.690	-0.079	0.623	0.334	0.033	0.256	0.106
SDNNIndex	-0.003	0.987	-0.088	0.584	-0.056	0.726	0.073	0.651	0.069	0.670
RMSSD	-0.108	0.502	0.019	0.907	-0.056	0.729	0.135	0.401	0.120	0.456
SDSD	-0.105	0.513	0.023	0.888	-0.052	0.748	0.143	0.371	0.125	0.435
NN50	0.126	0.432	-0.050	0.757	0.048	0.766	-0.086	0.592	-0.024	0.882
pNN50	-0.192	0.230	0.042	0.794	-0.094	0.560	-0.051	0.750	-0.100	0.535
TR.INDEX	0.062	0.698	-0.023	0.885	0.025	0.879	0.098	0.540	-0.218	0.171
ULF	-0.139	0.388	-0.154	0.337	-0.182	0.254	-0.162	0.312	-0.064	0.693
VLF	0.113	0.481	0.078	0.627	0.119	0.457	0.220	0.167	0.168	0.294
LF	-0.094	0.560	0.033	0.839	-0.038	0.812	0.241	0.129	0.227	0.153
HF	-0.077	0.631	0.058	0.719	-0.012	0.939	0.130	0.419	0.229	0.150
TP	-0.060	0.711	0.055	0.733	-0.003	0.985	0.191	0.232	0.242	0.127
LF/HF	-0.001	0.993	-0.231	0.147	-0.144	0.368	-0.071	0.659	-0.171	0.285

Pearson correlation
MinHR: Minimum heart rate beats/min, MaxHR: Maximum heart rate beats/min, AVGHR: Mean heart rate beats/min, NN: Cycle length between two normal beats, SDN: Standard deviation of all NN intervals during the examination, SDNN index: Mean of the standard deviations of all NN intervals in 5-min recordings, SDANN: Standard deviation of average NN intervals in 5-min recordings, NN50: Number of adjacent NN intervals with a difference of more than 50ms during the entire recording, pNN50: NN50 number divided by the total number of NNs, RMSSD: Square root of the sum of the differences of consecutive NN intervals in the 24-hour recording, TR.INDEX: Division of all NN interval number to the number of NN intervals in the mode length, HF: High-frequency band, LF: Low-frequency band, VLF: Very-low-frequency band, TP: Total power; variance of all NN intervals, ULF: Asleep, power in the low-frequency range

Discussion

Although many theories have been proposed to explain the aetiology of stuttering, our study focused on investigating the background of speech initiation. HRV parameters were examined in school-age children who stutter to examine possible differences in autonomic nervous system and these were compared with the values for children of similar age without any speech problems.

Our study found that the mean age in the stuttering group was 10.17 ± 2.75 years, which was not significantly different from the control group's mean age of 10.78 ± 2.78 years. This provided an advantage when evaluating the parameters. Karabulut (12) reported significant positive correlations with age for SDNN, SDNN-index, SDANN-index, low-frequency band (LF), very-low-frequency band, and total power parameters in their study of 51 healthy children with a mean age of 10.78 years. The emergence of different correlations in adults and children was attributed to the incomplete development of the autonomic nervous system in children (12). Previous studies found a negative correlation between HRV parameters and age in adults, while a significant positive correlation was found in children (13). In our study, no significant difference was found between age and HRV parameters in the case group.

In a study of 106 healthy children aged 1-20 years, Silveti et al. (14) found that SDNN and SDANN values were higher in boys, while SDNN-index, square root of the sum of the differences of consecutive NN intervals in the 24-hour recording (RMSSD), and pNN50 values did not differ between the genders. Similarly, Karabulut (12) reported no significant difference in HRV parameters between the genders in a study of 51 healthy children aged 4-17 years. Our study also found no significant difference in HRV parameters between genders.

Doruk et al. (2) investigated the presence of autonomic nervous system dysregulation in young adult stutters by analysing their HRV using a 24-hour Holter monitor. Compared to the control group, the stuttering group exhibited high RMSSD and differences in LF, LF/high-frequency band (HF), normalised LF, pNN50, SDNN, HF, normalised HF, total power, and GSI values. Additionally, negative correlations were found between subjective and total anxiety scores and LF and total power. Their study concluded that stuttering patients tended to have parasympathetic system dominance (2). Based on an analysis of our study results, the SDANN values of those children who stutter were significantly higher than those of the control group ($p=0.028$). No statistically significant

differences were observed between the groups in terms of the other variables ($p>0.05$). In our study, a weak positive correlation was identified between secondary behaviour values and SDANN values, a weak negative correlation with maxHR values, and a moderate negative correlation with Beats values ($p<0.05$).

SDANN is the standard deviation of the average NN intervals over short periods of 5 minutes. There are differing opinions regarding the SDANN parameter as a reflection of the autonomic nervous system. Fantoni et al. (15) interpreted an increase in SDANN as reflecting changes in autonomic tone, particularly changes in the interaction between the sympathetic and parasympathetic nervous systems in the heart. Adamson et al. (16) reported that a decrease in physical activity corresponds to a decrease in SDANN before clinical deterioration. Raj et al. (17) suggested that the change in SDANN value may not indicate a change in the autonomic nervous system, despite all these findings. The evidence supporting a positive correlation between the SDANN value and secondary behaviours in stuttering children suggests that physical activity may increase the SDANN value. Therefore, the significant SDANN value found in our study is interpreted as being related to the autonomic nervous system and secondary movements during speech in stuttering children. However, the lack of significant changes in the other HRV parameters does not support our hypothesis that the autonomic functions of stuttering children were different. In order to analyse the relationship between the autonomic nervous system and stuttering, it would be useful to use the autonomic nervous system in combination with other nervous dysfunction assessment tools.

To the best of our knowledge, there is no study in the literature investigating the relationship between stuttering and autonomic nervous system functions in school-age children using HRV measurements.

Study Limitations

There is the possibility of the population not being fully represented by both groups in our study. It would be beneficial to increase the number of participants in both groups and to work with more similar groups in terms of gender and age.

Conclusion

No significant correlation was found, except for the SDANN parameter, when examining the HRV parameters of school-age children with and without stuttering. The literature lacks sufficient and precise information regarding

the ability of SDANN to show autonomic function alone, which does not support our hypothesis that the autonomic functions of stuttering children are different. When examining the demographic characteristics (gender, age, and family history) of the group with stuttering, a positive and significant correlation was found between the SDANN parameter and secondary behaviours ($p>0.05$). However, there were no significant correlations between the other parameters, including stuttering severity, speech spontaneity, and the HRV parameters. The literature suggests that physical activity increases the SDANN value and supports a positive correlation with the SDANN value of secondary behaviours in stuttering children. This is the first study to examine autonomic dysfunctions of stuttering children using the 24-hour Holter measurement HRV. Our study may contribute to future research into the aetiology of stuttering.

Ethics

Ethics Committee Approval: The Clinical Research Ethics Committee of Ege University Faculty of Medicine approved this study (approval no.: 16-9/1, date: 14.10.2016).

Informed Consent: All patients received an Informed Consent Form, Case Report Form, and a hearing assessment with pure tone threshold audiometry.

Authorship Contributions

Surgical and Medical Practices: E.D., M.Ş., Z.Ü.T., M.F.Ö., Concept: P.Ö.U., M.Ş., M.F.Ö., Design: P.Ö.U., M.Ş., M.F.Ö., Data Collection or Processing: P.Ö.U., E.D., Z.Ü.T., Analysis or Interpretation: P.Ö.U., E.D., M.Ş., Z.Ü.T., M.F.Ö., Literature Search: P.Ö.U., E.D., M.Ş., Writing: P.Ö.U., E.D., M.Ş., Z.Ü.T., M.F.Ö.

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