



Autonomic activity in an adolescent with a single ventricle who underwent physical therapy intervention: case report

Atividade autonômica em uma adolescente com ventrículo único submetida à intervenção fisioterapêutica: relato de caso

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Abstract

Objective: To evaluate the autonomic activity of an adolescent with a single ventricle without surgical correction who participated in a physical therapy program. **Methods:** This was a 14-year-old female patient with a left-type single ventricle who had not undergone any surgical intervention. The autonomic activity was evaluated according to the heart rate variability in the supine and seated positions and by means of the respiratory sinus arrhythmia accentuation maneuver (RSA-M) in the beginning of the first, second and third years of outpatient cardiovascular physical therapy treatment. The RR intervals and beat-to-beat heart rate were calculated and stored for subsequent analysis. The heart rate variability was evaluated by means of the RMSSD and RMSM indexes and in the frequency domain with high and low frequency bands in normalized units (HFnu and LFnu, respectively) and using the LF/HF ratio. The cardiovascular physical therapy consisted of respiratory exercises associated with active and general resistance exercises, for two years. **Results:** From the first to the third year, the RMSM decreased (14.5%), LFnu band increased (42.2%), LF/HF ratio increased (117.0%) and HFnu decreased (35.2%). In all of the situations, the LFnu band was higher and HFnu was lower in the seated position. Additionally, the parasympathetic response to RSA-M increased from the first to the third years in 7.4% and 47.3%, respectively. **Conclusions:** We concluded that, for the patient studied, the reduction in heart rate variability seemed to be associated with the advance of the disease. Nevertheless, there was no damages related to postural change. In addition, the cardiovascular physical therapy proposed increased the parasympathetic response during RSA-M.

Key words: congenital cardiopathy; single ventricle; univentricular heart; autonomic nervous system; physical therapy.

Resumo

Objetivo: Avaliar a atividade autonômica de uma adolescente com ventrículo único (VU), sem correção cirúrgica, participante de um programa de fisioterapia. **Materiais e métodos:** Paciente do sexo feminino, 14 anos, com diagnóstico de VU tipo esquerdo, sem correção cirúrgica. A atividade autonômica foi avaliada pela variabilidade da frequência cardíaca (VFC) nas posições supina e sentada, e pela manobra para acentuar a arritmia sinusal respiratória (M-ASR) no início do primeiro (A1), segundo (A2) e terceiro anos (A3) de tratamento fisioterapêutico cardiovascular (TFC) ambulatorial. Os intervalos RR e a frequência cardíaca batimento a batimento foram calculados e armazenados para posterior análise. A VFC foi avaliada pelos índices RMSSD e RMSM e no domínio da frequência pelas bandas de alta e baixa frequência em unidades normalizadas (AFun e BFun, respectivamente) e pela razão BF/AF. O TFC constou de exercícios respiratórios associados a exercícios ativos e resistidos gerais, durante dois anos. **Resultados:** De A1 para A3, o RMSM reduziu (14,5%), a banda de BFun (42,2%) e a razão BF/AF aumentou (117,0%), e a banda AFun diminuiu (35,2%). Em todas as situações, a banda BFun foi maior e a AFun foi menor na posição sentada. Além disso, a resposta parassimpática à M-ASR aumentou da situação A1 para A3 em 7,4 e 47,3%, respectivamente. **Conclusões:** Concluímos que, na paciente estudada, a redução da VFC parece estar associada com o avanço da doença, porém, não houve prejuízos frente à mudança postural. Adicionalmente, o TFA proposto incrementou a resposta parassimpática durante a M-ASR.

Palavras-chave: cardiopatia congênita; ventrículo único; coração univentricular; sistema nervoso autônomo; fisioterapia.

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Introduction : : : .

Amongst the main congenital cardiopathies is the so-called single ventricle (SV) or univentricular heart, which can be classified into left ventricle, morphologically unique, where the single left or right ventricle predominates¹. The SV is usually associated with the transposition of the great vessels and to pulmonary stenosis, and the most effective treatment is surgery, carried out no later than in the first year of life. In case this correction does not occur, the average life expectancy has been of around 14 years².

In general, congenital cardiopathies alter the autonomic function, besides harming the vestibulosympathetic reflex function in the sinus node⁴. The evaluation of the heart rate variability (HRV) has been a method often used aiming at evaluating the cardiovascular risk, morbi-mortality, both in cardiovascular disorders⁴ as well as in conditions that might cause chronic hypoxemia⁵.

There are several factors that might affect HRV, among them respiratory cycles. During inhaling, heart rate (HR) tends to augment due to the vagal, while during exhaling there occurs the vagal retake and, therefore, HR tends to decrease. In this context, it is possible, by means of the Respiratory Sinus Arrhythmia Maneuver (RSA-M) to evaluate the integrity of the cardiorespiratory system⁶. Thus, the goal of the present case study was to prospectively evaluate HRV at rest and during RSA-M in an adolescent suffering from left-type SV without surgical correction, who took part in a out patient clinic program of cardiovascular physical therapy treatment (CVPTT).

Methods : : : .

Case report: Female patient, student, 14 years old, 1.63 m tall, 52kg, black-skinned, no previous cardiovascular surgery. At 3 months of age, the congenital anomaly was detected, after her mother reported that the child showed respiratory difficulties and intense perspiration during breast-feeding. After heart catheterism, left-type SV was diagnosed, as well as a moderate pulmonary stenosis, severe pulmonary hypertension, and transposition of the great vessels. The procedure was conservative, since the patient already demonstrated a severe pulmonary hypertension, indicative only of a heart-lung transplant. The patient started her treatment in the Cardiovascular Physical Therapy Sector at 12 and, during the physical therapy evaluation, she complained about strong lower-back pain and dyspnea after minor physical efforts (Functional Class III according to the New York Heart Association⁷); the saturation of peripheral oxygen (SpO₂) was at 80% at rest and the echo cardiogram indicated a preserved

ejection fraction. In the full blood exam, the values found for the red blood cells and for hemoglobin were above the reference values (actual values: 7,35 and 17,4; reference values: 4,20 a 5,50 million/mm³ and 12 to 16 gr/dl, respectively), and in the arterial blood gas analyses the values for PaO₂, PaCO₂ and arterial oxygen saturation were below the reference values (actual values: 44,40; 27,80 and 79,9; reference values: 75 to 90 mmHg, 30 to 42 mmHg and >90% respectively). The patient was making use of the following medications: anticonceptive (Desogestrel - 75 mg) once a day, and platelet anti-aggregant (Pentoxifilina - 400 mg) twice a day. The patient was informed about the experimental procedures and her guardian signed a term of free consent for participation form. The research was approved by the Universidade Federal de São Carlos Committee for Ethics in Research (protocol n. 065/2002).

The heart rate and the R-R interval registers (iR-R) were taken at rest, with the subject seated and in a supine position, during M-ASR, at the beginning of the first (A1), second (A2), and third (A3) years of the physical therapy intervention. The patient was advised to eat only light meals on the test days, and not to perform strenuous exercises or ingest stimulants (coffee, tea, sodas, etc) 24 hours prior to data collection. All procedures were carried out in the afternoon, so that the circadian cycle was respected. Room temperature and air humidity were kept between 22 and 24°C, and from 50 to 60%, respectively.

The electrocardiogram (ECG) signals, iR-R and HR were obtained on a beat-by-beat basis by means of a heart monitor (ECAFIX, TC 500, São Paulo, Brazil). For monitoring, the MC5 derivation was used. The register was recorded for 15 minutes at rest in a supine position and for 15 minutes while sitting. Later on, the ECG signals were transferred into a microcomputer and the analog signals were converted into binary values and then processed by a computer.

The Heart Rate Variability was analyzed in the time domain by the RMSM and RMSSD indexes. The RMSSD index corresponds to the square root of the difference in the sum of squares between iR-R on the record, divided by the determined time minus one. The RMSM, in turn, corresponds to the square root of the differences in the sum of squares of the individual values in relation to the mean, divided by the iR-R number at a given time. For the analyses of the signal in the frequency domain we used low (LFun= 0.04 a 0.15 Hz) and high frequencies (HFun= 0.15 a 0.4 Hz) in normalized units, and with the ratio BF/AF by means of Fast Fourier Transformation (FFT) of the data for the time series⁴, by means of a specific routine developed in the MatLab applicative (version 6.1.450 Release 12.1). The selection of the fragment to be analyzed – at rest, both in the supine and sitting positions, was made by means of the visual inspection of the iR-R

distribution over a 15-minute period, from which we selected the period characterized by the greatest signal stability and which showed at least 256 data points as was standardized by the *Task Force of the European Society of Cardiology and the North American Society of Pacing and Eletrophysiology*⁴.

The HR and iR-R recording during M-RSA were carried out for four minutes with the same equipment used for data collection at rest. Throughout the data recording, the patient was advised to inhale and exhale slowly and deeply several times, varying the pulmonary volume from pulmonary capacity to residual volume. The patient was told to inhale for five seconds and to exhale for five seconds, totaling six respiratory cycles per minute, through which we expected to obtain the maximal respiratory sinus arrhythmia⁶.

Next, the following indices for HR and iR-R were calculated, obtained from the M-RSA⁶: inhale/exhale ratio (I/E R); the average of the highest iR-R obtained during the expiratory phase divided by the average of the lowest iR-R in the M-RSA inhaling phase; and the inhale/exhale difference (Δ IE); and the difference between the highest HR average obtained during inhaling and the average of the lowest HR values obtained during the M-RSA expiratory phase.

The patient was attended to for one hour, twice a week during two consecutive years at the Cardiovascular Physical Therapy Sector of the Health Unit of the Universidade Federal de São Carlos. Her blood pressure (BP), HR, and SpO₂ were measured by means of a heart monitor (Omni 200, Minas Gerais, Brasil) before, during, and after the sessions. All exercises, consisting of series of 10 repetitions, were performed three times with one-minute intervals between them, namely: active and passive, upper, lower limb and trunk, free active exercises involving the trunk, shoulders, hips and strengthening of the upper limbs with barbells and 1kg ankle weights for the gluteal muscles, abdominal and paravertebral exercises. As the patient performed the exercises, she was encouraged to make use of diaphragmatic breathing.

Results

In regards to the time domain indices, no differences were observed in the annual evolution of the RMSSD index. However, RMSM demonstrated a reduction when comparing A1 with A2 (3%), A2 with A3 (12%), and A1 with A3 (15%). As for DF data, no differences were observed in the LFun or HFun bands from A1 to A2. Nonetheless, there was a LFun increase (49 and 42% respectively) and an HFun reduction (34 and 35% respectively) from A2 to A3 and from A1 to A3. Likewise, the BF/AF ratio indicated an increase (131%) from situation A2 to A3 and from A1 to A3 (117%), although differences were not observed from A1 to A2. It has been observed that in A1, A2, and A3, LFun was greater (69, 57 and 23%, respectively), and HFun was smaller (54.77 and 75%, respectively) in the sitting position when compared to the supine position. Besides this, one could observe that I/ER increased from A1 to A2 (2%), from A2 to A3 (5%), and from A1 to A3 (7%). For Δ IE, an increase was observed from A2 to A3 (25%), as well as from A1 to A3 (47%) (Table 1).

Discussion

The patient under study demonstrated an HRC reduction between the prospective evaluations. This fact can be attributed to the degree of hypoxemia present, which may produce deleterious effects on the autonomic responses^{5,9}. The present results showed that the HRC changes *vis-à-vis* hypoxemia remained steady during the treatment period (79 and 81 and 80% for A1, A2 e A3, respectively) and the values for blood pressure and HR did not suffer any changes, whatsoever. Another factor to be taken into account was the possible progression of the disease, since the patient under study was already reaching the mean life expectancy age.² In this aspect, according to Massin et al.¹⁰, the greater the congenital cardiopathy, the higher is the degree of HRC harm.

Table 1. Heart rate variability values of the studied patient, in the time and frequency domains, during the evaluations in the supine position and with postural changes (supine to sitting position), and the respiratory sinus arrhythmia accentuation maneuver indices.

	TD Supine		FD Supine			FD Seated		RSA-M Supine	
	RMSSD (ms)	RMSM (ms)	LF nu	HF nu	LF/HF	LF nu	HF nu	E/IR	Δ IE
A1	19.54	27.78	0.45	0.54	0.82	0.76	0.23	1.11	10.03
A2	18.92	21.89	0.43	0.53	0.77	0.69	0.30	1.13	11.83
A3	16.69	19.70	0.64	0.35	1.78	0.79	0.20	1.19	14.77

TD: time domain; FD: frequency domain; RSA-M: respiratory sinus arrhythmia accentuation maneuver; A1: initial evaluation; A2: second evaluation; A3: third evaluation; RMSSD: corresponded to the square root of the sum of squares of the differences between the R-R intervals registered, divided by the number of R-R intervals (R-Ri) in a specified time period minus one; RMSM: corresponded to the square root of the sum of squares of the differences of the individual values in relation to the mean value, divided by the number of R-Ri for a specified time period; LF: low frequency; HF: high frequency; normalized units (nu); RE/I: mean of the highest values of the R-Ri obtained during the expiratory phase divided by the means of the lowest R-Ri of the inspiratory phase; Δ IE: differences between the highest mean values of the heart rate (HR) obtained during the inspiratory phase and mean of lowest HR values during expiratory phase of the RSA-M.

Concerning the M-RSA, the present results revealed that both E/IR and Δ IE showed greater values in the following evaluations. In this aspect, the respiratory sinus arrhythmia is a product of the interaction between the respiratory and the cardiovascular systems, since there is evidence that the variations in the current volume and HR can potentially interfere with vagal control⁶. Therefore, it is possible to infer that the cardiovascular physical therapy treatment applied did benefit the parasympathetic activity. These aspects are clinically important, since the interventions that increased the vagal control to

cardiovascular patients should be encouraged, contributing to the reduction of the incidence of morbi-mortality⁶.

We finally concluded that for the patient under study, type SV congenital cyanotic cardiopathy when not surgically corrected, produced a reduction of HRC with an increase of the sympathetic activity although without harming the adjustment *vis-à-vis* postural changes. The clinical cardiovascular physical therapy program put forward seems to have contributed to the increment of the parasympathetic response during the M-RSA.

References

1. Büchler JR. Ventrículo único: critério morfológico na classificação e conceituação das cardiopatias congênitas. *Arq Bras Cardiol.* 1981;36(4):225-6.
2. Atik E. Univentricular atrioventricular connection. Review of the current therapeutical possibilities. *Arq Bras Cardiol.* 2000;74(1):3-4.
3. Kirjavainen T, Viskari S, Pitkänen O, Jokinen E. Infants with univentricular heart have reduced heart rate and blood pressure responses to side motion and altered responses to head-up tilt. *J Appl Physiol.* 2005;98:518-25.
4. Task Force of the European Society of Cardiology and the North American Society of Pacing and Electrophysiology. Heart rate variability: standards of measurements, physiological interpretation, and clinical use. *Circulation.* 1996;93:1043-65.
5. Chen WL, Chen GY, Kuo CD. Hypoxemia and autonomic nervous dysfunction in patients with chronic obstructive pulmonary disease. *Respir Med.* 2006;100:1547-53.
6. Yasuma F, Hayano J. Respiratory sinus arrhythmia. Why does the heartbeat synchronize with respiratory rhythm? *Chest.* 2004;125:683-90.
7. American College of Cardiology/American Heart Association Task Force on Assessment of Cardiovascular Procedures (Subcommittee on Exercise Testing). Exercise Testing Task Force Members: Guidelines for Exercise Testing. *J Am Coll Cardiol.* 1986;8(3):725-38.
8. Silva E, Catai AM, Trevelin LC, Guimarães JO, Silva Jr LP, Silva LMP, et al. Design of a computerized system to evaluate the cardiac function during dynamic exercise. *Anais do World Congress of Medical Physics and Biomedical Engineering* 1994. p409.
9. Stewart AG, Waterhouse JC, Howard P. The QTc interval, autonomic neuropathy and mortality in hypoxaemic COPD. *Respir Med.* 1995;89:79-84.
10. Massin M, Von Bernuth G. Clinical and haemodynamic correlates of heart rate variability in children with congenital disease. *Eur J Pediatr.* 1998;157:967-71.