Progress in Pediatric Cardiology xxx (xxxx) xxx–xxx

Contents lists available at ScienceDirect

Progress in Pediatric Cardiology

journal homepage: www.elsevier.com/locate/ppedcard

Can a home-based cardiac physical activity program improve and sustain quality of life and exercise capacity in children with Fontan circulation?☆

Roni Jacobsen[a,*,1,2], Michael Danduran[b,c], Kathleen Mussatto[b], Garick D. Hill[a,3], Salil Ginde[a,d]

[a] Department of Pediatrics, Division of Pediatric Cardiology, Medical College of Wisconsin, 9000 W. Wisconsin Avenue, MS 713, Milwaukee, WI 53226, United States of America
[b] Department of Pediatrics, Division of Pediatric Cardiology, Children's Hospital of Wisconsin, 9000 W. Wisconsin Avenue, MS 713, Milwaukee, WI 53226, United States of America
[c] College of Health Sciences, Program in Exercise Science, Marquette University, PO Box 1881, Milwaukee, WI 53201-1881, United States of America
[d] Department of Internal Medicine, Division of Cardiovascular Medicine, Medical College of Wisconsin, 9000 W. Wisconsin Avenue, Milwaukee, WI 53226, United States of America

ARTICLE INFO

Keywords:
Congenital Heart Disease, CHD
Fontan
Exercise
Physical activity
Rehabilitation
Quality of life

ABSTRACT

Objective: Fontan survivors report reduced health-related quality of life (HRQOL) and have decreased exercise tolerance compared to healthy peers. We recently demonstrated that a 12-week home-based physical activity program was safe, feasible, and improved parent proxy-reported HRQOL and objective measures of exercise capacity in patients with Fontan circulation. We sought to determine if these improvements in HRQOL and exercise capacity are sustained at 6-month follow-up.

Design: Patients, 8–12 years old, with Fontan circulation that completed a 12-week moderate-to-vigorous intensity home-based physical activity program were invited to attend a 6-month follow-up session to complete objective assessments of HRQOL and exercise capacity. HRQOL was measured with validated questionnaires. The 20-meter Shuttle Run (PACER Test) was used to measure exercise capacity.

Results: Of the 13 patients who completed the original 12-week physical activity program, 11 (85%) attended the 6-month follow-up. There were no adverse events during the follow-up period. Improvements in parent proxy-report for HRQOL at completion of the 12-week program were sustained at 6-month follow-up. Patients reported a significant decrease in HRQOL assessed by PedsQL at 6-month follow-up. However, there was a non-significant trend of improvement in patient PCQLI total and psychosocial impact scores at completion of the 12-week program, which was sustained at 6-month follow-up. The significant improvements in objective measures of exercise capacity seen from baseline to completion of the 12-week program were sustained at 6-month follow-up.

Conclusion: Improvements in parent-proxy report HRQOL and objective measures of exercise capacity after completion of a home-based 12-week physical activity program were sustained at 6-month follow-up. Despite improvements in exercise capacity, patients did not report improved HRQOL. A larger, controlled study of this home-based physical activity program is needed to assess the impact of physical activity and improved exercise capacity on HRQOL in children with Fontan circulation.

☆ Declarations of interest: None.

* Corresponding author at: 13123 E 16th Street, B100, Aurora, CO 80045, United States of America.
E-mail address: roni.jacobsen@childrenscolorado.org (R. Jacobsen).
[1] Present address: Department of Pediatrics, Division of Pediatric Cardiology, University of Colorado School of Medicine, 13123 E 16th Street, B100, Aurora, CO 80045, United States of America.
[2] Present address: Department of Internal Medicine, Division of Cardiology, University of Colorado School of Medicine, 12605 E 16th Avenue, Aurora, CO 80045, United States of America.
[3] Present address: Department of Pediatrics, Division of Pediatric Cardiology, Cincinnati Children's Hospital Medical Center, 3333 Burnet Avenue, Cincinnati, OH 45229, United States of America.

https://doi.org/10.1016/j.ppedcard.2018.06.003
Received 6 October 2017; Received in revised form 29 June 2018; Accepted 29 June 2018

1. Introduction

Several studies have demonstrated decreased health-related quality of life (HRQOL) and exercise capacity in children with congenital heart disease (CHD). Patients with CHD also have lower levels of daily physical inactivity and an increased prevalence of obesity and other cardiovascular risk factors, compared to healthy children [1]. These findings are likely due to a combination of a national trend in increased sedentary behaviors, physician imposed restrictions, parental barriers, and impaired exercise tolerance in children with CHD. Regular physical activity not only improves fitness but can also have positive effects on school performance, improved social relationships, and improved mental health. Current guidelines exist to promote increased physical activity in healthy children, recommending at least 30–60 min of daily moderate-to-vigorous exercise or physical activity [1–3]. The American Heart Association recently published a statement which specifically promotes regular physical activity in patients with CHD [1].

Physical activity programs improve measures of health-related quality of life (HRQOL) as well as exercise capacity in pediatric patients with CHD [4–7]. In a recent pilot study, we demonstrated participation in a moderate-to-high intensity home-based cardiac physical activity program was safe and feasible in 13 patients 8–12 years-old with Fontan circulation. In addition, we found these patients had significant improvements in parent-proxy reported HRQOL and objective measures of exercise capacity [7]. However, the longer term benefits of participation in a 12-week program are unknown. We sought to determine whether the improvements seen in HRQOL and exercise capacity following completion of an innovative 12-week home-based cardiac physical activity program were sustained at 6-month follow-up.

2. Methods

The study was approved by the Children's Hospital of Wisconsin Internal Human Research Review Board and was conducted in accordance with all human research regulatory requirements. Informed assent of the study participant and consent of the parent or guardian were obtained, according to institutional guidelines. Data from the initial pilot study have been previously described [7].

2.1. Subjects

We asked 13 patients, 8–12 years old, with Fontan circulation that had completed a 12-week moderate-to-vigorous intensity interval training home-based cardiac physical activity program to return for a 6-month in-person follow-up visit to undergo repeat assessments of HRQOL and exercise capacity.

2.2. Physical activity program

As previously described, the 12-week home-based moderate-to-high intensity physical activity program consisted of 3 primary components: (1) a 45-minute home exercise routine of dynamic and static exercises, depicted on DVD or paper handout, for patients to complete 3 to 4 times per week and (2) 3 formalized in-person exercise sessions, and (3) scheduled telephone calls with a college student mentor [7]. Additional activity during the study period was not limited and patients were encouraged to be as active as desired. Upon completion of the 12-week study, patients were encouraged to continue to participate in higher levels of physical activity.

At 6-month follow-up, patients and their parents completed the previously validated Pediatric Quality of Life Inventory Generic 4.0 Score (PedsQL)© and Pediatric Cardiac Quality of Life Inventory (PCQLI)© questionnaires to assess physical function HRQOL. The PCQLI© is a disease-specific HRQOL instrument for children (8–12 years of age) with congenital or acquired heart disease. The PedsQL and PCQLI forms include both self-respondent and parent proxy reporting [8–12]. Exercise capacity was assessed with exercise duration and estimates of maximal oxygen uptake (VO2max) based on performance on the 20-meter Shuttle Test Run© [13–16].

2.2.1. Data outcomes and risk factors

Each patient's height, weight, and BMI were recorded at the 6-month follow-up. In addition, each patient's most recent echocardiogram was reviewed and the systemic ventricular systolic function assessed semi-quantitatively by a single pediatric cardiologist. The resting and immediately post Shuttle Test Run oxygen saturation, blood pressure, and heart rate were recorded for each patient. Exercise time and number of shuttles completed were recorded. VO2 was calculated based on the patient's gender, age, BMI, and total number of shuttles (TL) completed, using the validated equation from Matsuzaka et al. [17].

Results from the 6-month follow-up in-person session were compared to the data obtained at the baseline session, prior to intervention, as well as at completion of the 12-week physical activity program.

3. Statistical analysis

Descriptive statistics, mean with standard deviation or median with ranges, were used to summarize the demographic characteristics of study subjects. The primary outcome measure in our study was the physical function HRQOL, measured with the PedsQL and PCQLI scores, calculated at baseline, immediately after the 12-week intervention, and then 6 months after completion of the program. The secondary outcome measure was change in exercise capacity as measured by shuttle number, time, and distance, and the calculated VO2 max assessed with the Shuttle Test Run during the same time periods. Other physical parameters of height, weight, BMI, and resting and immediately post-exercise heart rate, blood pressure, and oxygen saturation were also analyzed.

Physiologic parameters are presented as mean with standard deviation. Paired sample two-tailed Student *t*-tests were performed to compare differences between pre- and post- intervention outcomes and also post-intervention and at 6-month follow-up. Questionnaire data was scored and totaled per published instrument standards. Total scores from the questionnaires were compared post 12-week intervention to 6-month follow-up using Student *t*-tests. If violations of parametric data occurred, non-parametric assessment of differences in post-intervention and follow-up values were calculated using the Wilcoxin Sign rank test. Data analysis was performed using StatalC13 (StataCorp, College Station, Texas) with statistical significance established at $P < 0.05$.

4. Results

Of the 13 patients who completed the original 12-week physical activity program, 11 (85%) attended the 6-month in-person follow-up session. The patient characteristics are shown in Table 1. The median

Table 1
Demographics and baseline characteristics.
This table shows the patient characteristics of the 11 patients who attended the 6-month follow-up session.

Patient characteristics (N = 11)		
Variable	Number (%)	Range
Mean age	10.5 yrs.	9–12 yrs.
Time since Fontan	7.4 ± 2.3 yrs.	1.7–11.1 yrs.
Females	6 (55%)	
Dominant ventricle		
RV	6 (55%)	
LV	5 (45%)	
Fenestration	5 (45%)	
Normal cardiac function	11 (100%)	
Pacemaker/ICD	0 (0%)	