

The Effects of Walking Combined with Active Range of Motion Exercises on Aerobic Capacity in Patients with Juvenile Dermatomyositis

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Abstract

Objectives: Physical activity programs may be useful in the treatment of Juvenile Dermatomyositis (JDM). Therefore, we aimed to assess the effects of combined physical activity program on aerobic capacity, life quality and function in children with JDM.

Methods: 27 children with JDM and 15 healthy children as a control group were included. In physical activity program, all patients performed both aerobic walking exercise (3 days/week/12 weeks) and active range of motion exercises (4 days/week/12 weeks). All patients completed the childhood health assessment questionnaire (CHAQ). Range of motions (ROM) measurements of joints were performed by using universal goniometer. Aerobic capacity was determined by measuring peak oxygen uptake (VO₂peak) during a cardiopulmonary exercise testing (CPET).

Results: Before and after physical activity program peak oxygen uptake were significantly lower in patient group than in control group. After physical activity program, certain cardiopulmonary exercise testing parameters of JDM patients significantly improved. Peak oxygen uptake was found to be significantly increased following physical activity program in JDM patients. Also time-to-exhaustion was significantly increased after physical activity program. Physical activity program significantly improved CHAQ score in patient group.

Conclusions: As well as improving life quality and function, aerobic walking combined with active range of motion exercises improve aerobic capacity of patients with JDM.

Keywords: Juvenile dermatomyositis; Physical activity; Aerobic capacity; Peak oxygen uptake; Range of motion; Exercise; Childhood health assessment questionnaire

Introduction

Juvenile dermatomyositis (JDM) is an idiopathic inflammatory myopathy and commonly presents with proximal muscles weakness and nonsuppurative inflammation of the skeletal muscle. It is the most common idiopathic nonsuppurative inflammatory myositis of children and adolescents [1,2]. It is a multisystemic disease that may involve joints, lungs, heart and other internal organs, especially skin and striated muscles [3,4]. However, myocardium, lung, and gastrointestinal involvements are rarely seen [5,6]. Children with JDM may experience difficulty in voluntary movements and they generally show a positive Gower's sign. These patients may have arthritis and flexion contractures may develop in the joints that are involved [7,8].

In the past years physicians were avoiding to recommend physical exercises to JDM patients as they believe that physical activity could increase muscle inflammation or activity of the disease [9]. However, the growing numbers of evidence suggest beneficial effects of physical activity in JDM. In a study it was shown that a single session of resistance exercise neither increases inflammation nor leads a decrease in muscle functions in patients with JDM [10]. In a study improved

muscle strength and function was reported after a 12-week supervised exercise training program. Also this training program provided improvements in bone mass, muscle mass, health-related quality of life and aerobic exercise capacity in JDM patients [9]. In a patient 16 weeks of combined aerobic and resistance exercise program lead an increase in the aerobic capacity and muscle functions [11].

Therefore, physical exercises may be a useful treatment option in the management of JDM in means of increasing range of motion (ROM), muscle strength and neuromuscular coordination. Also it may help in increasing the functional capacity of these patients. In the present study we aimed to investigate the effects of physical activity program (combined walking and active ROM exercises) on aerobic capacity, ROM in certain joints, and quality of life in patients with JDM.

Patients and Methods

Subjects

27 children (17 female, 10 male) with chronically stable disease phase of juvenile JDM were included in the study. 15 healthy children (9 female, 6 male) served as controls. Controls did not participate in any regular sportive activities. All of the patients were diagnosed as JDM according to the Bohan and Peter criteria by pediatric rheumatologists (OK and KB) [12,13]. Exclusion criteria were a) severe

heart or lung conditions b) chronic musculoskeletal disorder c) not being able to exercise. The Turkish version of childhood health assessment questionnaire (CHAQ) was completed by the parents or the guards of children after giving information and answering the questions raised by parents [14]. All parents of subjects gave informed consent to participate in the study, which was approved by the local ethics committees at Istanbul University. The study followed the World Medical Association's 2013 Declaration of Helsinki.

Anthropometry and ROM measurements

Body mass was measured on a balance beam medical scale (Fairbanks) to the nearest 0.1 kg. Stature was measured on a stadiometer (Holtain, UK) to an accuracy of ± 0.5 cm with the subject bare foot, feet together, and head level. ROM values in shoulder, back, hip, and knee joints were measured; triple and average results were recorded. For normal ROM values of joints, Kendall-McCreary norms were used [15]. Body composition analyze was performed by analyzer (TANITA, Type TBF-300 M).

Experimental protocol

The peak oxygen uptakes (VO_{2peak}) of all subjects were measured during a cardiopulmonary exercise testing (CPET). Each child first underwent a comprehensive physical examination which included a 12-lead electrocardiogram (ECG) recording and blood pressure measurement at rest. Prior to exercise tests, all participants underwent pulmonary function tests (Ocean Win Spiro 2.36 B software with Spirobank; Medical International Research, Italy). All tests were performed in an air-conditioned laboratory room at 20-22°C and 40% relative humidity of air, to minimize thermal stress. Subjects had a light meal 2 hours before exercise and abstained from strenuous exercise for a week prior to test protocol.

Cardiopulmonary exercise testing

Whereas CPET was applied to control group to assess the baseline values only at the beginning of the study, it was applied to patient group before and after physical activity program. All the children were tested with Bruce exercise test protocol. Each test was terminated by peak exercise level. It was considered peak level if the subject achieved two of the three following test criteria: (1) the plateau of oxygen uptake with increasing work load, (2) a respiratory exchange ratio (RER) of 1.10 or higher, and (3) heart rate reaching 85% of age-predicted maximal heart rate (Max HR). This parameter was calculated by subtracting subject's age from 220 (Max HR=220 age). During CPET, 12-lead ECG was monitored. In all cases, a Quinton 5000 recorder and lead system (Quinton Instrument Company, Seattle, USA) were utilized to monitor and record ECG. Heart rate was monitored from ECG. Blood pressure was measured every stage of the Bruce protocol using cuff manometer for children (ERKA, Germany).

Oxygen uptake (VO_2), carbon dioxide production (VCO_2) and ventilation (VE), were monitored continuously, breath-by-breath, at rest, during CPET, and for 5 minutes of recovery after exercise, using a CPET system (Cortex MetaLyzer 3B, Cortex Biophysik GmbH, Leipzig, Germany). The system was calibrated before each test with standard gases of known O_2 and CO_2 concentrations. The most elevated VO_2 measured over 30 seconds during the last period of the CPET was considered as the VO_2 peak. RER was calculated as VCO_2/VO_2 . V-slope method was used to determine the anaerobic threshold (AT).

Physical activity program

A home activity program (walking combined with ROM exercises) was planned for patients. Prior to program detailed information about the program was given to both patients and their parents. Their compliance to program was routinely followed by phone calls.

Walking

Patients walked 3 days/week, for 12 weeks (outdoor walking). Program was started with 30 minutes walking during the first week. Then, the exercise duration was incremented 5 minutes for each two weeks. Walking intensity adjustment was based on AT determined by CPET for each patient to achieve a moderate intensity walking. Before and after the walking sessions, stretching exercises were performed for warming up and cooling down.

Active ROM exercises

Patients performed active ROM exercises as 10 times/set, 2 sets/day, 4 days/week, for 12 weeks. It included 1) Shoulder exercises: Shoulder movements (up and down, side to side); shoulder rotation (roll forward and back) 2) Spine exercises: Spine movements; flexion, extension, rotation and side bending 3) Hip exercises: hip and knee bends; leg lifts, leg movements (side to side) 4) Knee exercises: Knee movements (extension and flexion)

Statistical analysis

The SPSS version 13.0 statistic program was used for the statistical analyses. Physical characteristics and CPET results of juvenile JDM and control groups were compared using "Mann Whitney U-Test".

Changes in CHAQ scores and CPET results, as well as goniometric measurements before and after PA program were compared by using the "Wilcoxon Signed Ranks Test" in patients with Juvenile JDM. In all comparisons p values <0.05 were considered statistically significant.

Results

Physical characteristics of groups are given in Tables 1 and 2. The median age at the onset of diagnosis was 6.2 ± 4.1 years and mean follow up period was 65.2 ± 48.6 months. All patients had heliotrope rash and had elevated muscle enzymes at the disease onset. Gottron papule was inspected in 25 of patients (93%). 11 patients had calcinosis (41%). In the physical examination proximal muscle weakness was determined in 25 of patients (93%). All patients received corticosteroid treatment. Corticosteroids were combined with methotrexate (MTX) at the mean dosage of 13.6 ± 5.8 mg/week (5-60 mg/week). Patients nonresponsive to steroid and MTX were treated with additional cyclosporine therapy.

JDM and control groups were age matched and there was no significant difference in the physical characteristics between the groups before and after PA program (Table 1 and Table 2 respectively). In comparison of CPET results, there were significant differences in VO_{2peak} , VE peak, VO_{2AT} , and HRAT resting HR between the two groups before PA program ($p=0.039$, $p=0.027$, 0.045 , $p=0.003$ and 0.047 respectively; Table 3).

In addition, there were statistically significant differences in VO_2 peak, VE peak, HRAT, HR Rest between two groups after PA program ($p=0.041$, $p=0.027$, $p=0.007$ and $p=0.036$ respectively; Table 4). To determine the effects of PA program, we compared CHAQ score and

CPET results before and after completion of PA program. There were significant differences in CHAQ score, VO₂ peak, VE peak, time-to-exhaustion, VEAT (p=0.0001, p=0.004, p=0.004, p=0.001 and p=0.010 respectively; Table 5).

Comparison of ROM values before and after PA program revealed significant changes in left shoulder flexion, lateral flexion of spine to both right and left side, and right and left knee flexion were significantly changed (p=0.024, p=0.046, p=0.039, p=0.007 and p=0.006 respectively; Table 6).

Parameters	Patient group (n=27) Mean ± SD	Control group (n=15) Mean ± SD	P value
Age, year	11.59 ± 3.53	11.93 ± 2.63	0.712
Height, cm	146.56 ± 16.00	154.80 ± 15.79	0.103
Weight, kg	44.91 ± 16.73	50.44 ± 13.46	0.259
Body Mass Index, %	19.84 ± 4.09	20.65 ± 3.43	0.423
Basal Metabolic Rate, kcal	1312.37 ± 277.41	1439.40 ± 247.70	0.086
Fat Ratio, %	18.00 ± 8.26	18.95 ± 10.57	0.927
Fat Mass, kg	8.81 ± 5.78	10.23 ± 7.23	0.703

Table 1: Physical characteristics of groups before PA program

Parameters	Patient group (n=27) Mean ± SD	Control group (n=15) Mean ± SD	P value
Age, year	11.63 ± 3.51	11.93 ± 2.63	0.761
Height, cm	148.37 ± 15.70	157.00 ± 15.01	0.076
Weight, kg	45.30 ± 15.86	51.84 ± 12.93	0.212
Body Mass Index, %	19.83 ± 3.69	20.69 ± 3.37	0.351
Basal Metabolic Rate, kcal	1354.67 ± 256.43	1450.60 ± 249.16	0.16
Fat Ratio, %	18.04 ± 8.02	20.80 ± 9.62	0.609
Fat Mass, kg	9.86 ± 5.50	11.30 ± 7.22	0.948

Table 2: Physical characteristics of groups after PA program

Parameters	Patient group (n=27) Mean ± SD	Control group (n=15) Mean ± SD	P value
VO ₂ peak (ml/kg/min)	28.30 ± 5.25	33.40 ± 7.67	0.039
VE peak (L/min)	58.34 ± 15.49	75.01 ± 23.54	0.027
HR Max (bpm)	193.37 ± 10.09	194.40 ± 8.79	NS
time-to-exhaustion (sec)	551.30 ± 140.18	614.40 ± 159.10	NS
VO ₂ AT (ml/kg/min)	22.07 ± 4.35	25.80 ± 5.67	0.045

Parameters	Before (n=27) Mean ± SD	After (n=27) Mean ± SD	P value
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HRAT (bpm)	155.59 ± 12.27	167.13 ± 8.79	0.003
VEAT (L/min)	38.33 ± 12.08	45.30 ± 14.71	NS
HR Rest (bpm)	102.22 ± 12.99	92.40 ± 16.76	0.047

Table 3: CPET results of groups before PA program (Abbreviations: HR: heart rate, VE: minute ventilation, Max: maximal, AT: anaerobic threshold, Rest: Resting, bpm: beat per minute, sec: second, NS: not significant)

Parameters	Patient group (n=27) Mean ± SD	Control group (n=15) Mean ± SD	p value
VO ₂ peak (ml/kg/min)	29.96 ± 5.35	35.07 ± 7.85	0.041
VE peak (L/min)	62.48 ± 17.54	79.33 ± 23.18	0.027
HR Max (bpm)	192.81 ± 11.27	194.80 ± 8.35	0.732
time-to-exhaustion (sec)	599.11 ± 131.12	633.33 ± 156.66	0.713
VO ₂ AT (ml/kg/min)	23.00 ± 4.57	26.53 ± 7.57	0.055
HRAT (bpm)	156.41 ± 10.98	166.40 ± 8.93	0.007
VEAT (L/min)	40.05 ± 10.96	45.45 ± 15.91	0.454
HR Rest (bpm)	100.07 ± 13.43	89.73 ± 15.06	0.036

Table 4: CPET results of groups after PA program (Abbreviations: HR: heart rate, VE: minute ventilation, Max: maximal, AT: anaerobic threshold, Rest: Resting, bpm: beat per minute, sec: second)

Parameters	Before (n=27) Mean ± SD	After (n=27) Mean ± SD	P value
CHAQ score	0.60 ± 0.46	0.28 ± 0.27	0
VO ₂ peak (ml/kg/min)	28.30 ± 5.25	29.96 ± 5.35	0.004
VE peak (L/min)	58.34 ± 15.49	62.48 ± 17.54	0.004
HR Max (bpm)	193.37 ± 10.09	192.81 ± 11.27	NS
time-to-exhaustion (sec)	551.30 ± 140.18	599.11 ± 131.12	0.001
VO ₂ AT (ml/kg/min)	22.07 ± 4.35	23.00 ± 4.57	NS
HRAT (bpm)	155.59 ± 12.27	156.41 ± 10.98	NS
VEAT (L/min)	38.33 ± 12.08	40.05 ± 10.96	0.01
HR Rest (bpm)	102.22 ± 12.99	100.07 ± 13.43	NS

Table 5: CHAQ scores and CPET results of the patient group before and after PA program (Abbreviations: CHAQ: childhood health assessment questionnaire, VO₂: oxygen uptake VE: minute ventilation, HR: heart rate, Max: maximal, AT: anaerobic threshold, Rest: resting, bpm: beats per minute, sec: second, NS: not significant.)

Shoulder abduction	Right	148.27 ± 3.99	148.65 ± 3.62	NS
	Left	148.65 ± 3.02	149.42 ± 1.63	NS
Shoulder flexion	Right	148.73 ± 6.16	149.69 ± 5.14	NS
	Left	146.35 ± 6.41	149.23 ± 2.32	0.024
Spine flexion		87.50 ± 4.53	88.65 ± 3.33	NS
Spine extension		23.85 ± 2.03	24.42 ± 1.63	NS
Spine lateral flexion	Right side	24.04 ± 2.01	24.62 ± 0.21	0.046
	Left side	24.02 ± 1.89	24.81 ± 0.98	0.039
Hip flexion (with knee extension)	Right	99.04 ± 2.46	99.42 ± 2.16	NS
	Left	98.08 ± 4.26	98.85 ± 3.55	NS
Hip flexion (with knee flexion)	Right	98.65 ± 3.33	99.17 ± 2.54	NS
	Left	98.27 ± 4.23	99.23 ± 2.72	NS
Hip extension	Right	29.42 ± 1.63	30.00 ± 0.00	NS
	Left	29.31 ± 1.69	30.00 ± 0.00	NS
Knee flexion	Right	144.62 ± 7.47	149.20 ± 2.36	0.007
	Left	145.19 ± 7.00	148.85 ± 3.26	0.006
Knee extension	Right	0.00 ± 0.00	0.00 ± 0.00	NS
	Left	0.00 ± 0.00	0.00 ± 0.00	NS

Table 6: ROM results of the patient group before and after PA program (Abbreviations: NS: not significant)

Discussion and Conclusion

JDM is characterized by various severities of vasculitis which is observed early in disease course [2]. This vasculopathy principally affects muscles and skin. The weakness of the proximal muscles is pathognomonic clinical findings in patients with JDM [2]. It has been shown that JDM patients often experience low lean and bone mass, marked weakness, poor aerobic conditioning, and strong exercise intolerance [16-18]. Quality of life may be impaired compared with healthy controls [19] in both physical and psychosocial domains, requiring psychosocial support. Patients with Polymyositis (PM) and Dermatomyositis (DM) display fewer oxidative slow-twitch type I muscle fibers and lower aerobic capacity compared to healthy controls [20-22]. This could be another factor that contributes to muscle weakness in JDM.

In our study, in the CPET applied before the PA program, VO₂ peak, VO₂AT, VE peak, and HRAT levels of the JDM group were significantly lower than control group (p=0.039; p=0.045; p=0.027 and p=0.003 respectively; Table 3). In the same CPET HR rest of the JDM group was significantly higher than control group (p=0.047; Table 3). Depending on these findings cardiopulmonary exercise capacity of JDM group is lower than control group.

After PA program VO₂ peak, VE peak, and HRAT levels in JDM group were significantly lower than control group (p=0.041, p=0.027 and p=0.007, respectively; Table 4). HR rest of the JDM group was significantly higher than control group (p=0.036; Table 4). After PA

program cardiopulmonary exercise capacity of JDM group was lower than control group.

Beneficial effects of exercise and its safety has been reported in recent studies [23-25]. Also beneficial effects of regular exercise program have been shown in another idiopathic inflammatory disease such as juvenile idiopathic arthritis [26]. There are also studies revealing that exercise may protect these patients from muscle mass loss that occurs due to muscle inflammation, physical inactivity and glucocorticoid treatment [27,28].

In the present study, after PA program, certain CPET parameters of JDM patients significantly improved. VO₂ peak, VE peak, and VEAT were found to be significantly increased following PA program in patient group (p=0.004; p=0.004; p=0.01, respectively; Table 5). Time-to-exhaustion was also significantly increased after PA program (p=0.001; Table 5). These results reveal important cardiopulmonary adaptations to the PA program.

Recently, in a study, Omori et al. [9] applied a 12 weeks (2 day/week; 1 hour/day) of regular and combined (aerobic and resistant training) exercise program to JDM patients. Aerobic exercise (30 min walking) was performed on a motorized treadmill. In their study, exercise HR adjustment was based on 70% of VO₂ peak for each patient. After exercise program they found 13.3 % increase in VO₂ peak level, and 18.2% in time-to-exhaustion in JDM group. In the same study patients also performed resistance type exercises and this exercises lead significant improvements in the muscle strength of upper and lower extremities (p<0.05) [9].

The duration of the PA program in our study was 12 weeks as in this study. Unlike our study, exercise program was performed under supervision and aerobic exercises were combined with resistance type exercises instead of ROM exercises in this study. However, we found similar improvements in VO₂ peak and time-to-exhaustion. In another study Munters et al. [23] reported an increase in endurance exercise performance following a 12 weeks of endurance training program in patients with stable PM and DM. Also they observed a significant increase in VO₂ Max and time-to-exhaustion in these patients.

Non-destructive, sequel-free arthritis may occur during the active period of JDM. Arthritis is usually polyarthritis, and joint flexion contractures may develop [7,8]. Calcinosis is not a rare complication in JDM patients and it has important effects on the morbidity of the patients especially in means of restricting physical activity. However, this complication usually occurs at the later stages of the disease [29-33]. In the medical treatment of all patients in our study, corticosteroids and MTX were used in combination. Despite this aggressive therapy, calcinosis was detected in 11 (41%) patients in different regions of the body (arm, elbow, knee and hip). As well as muscle contractures, calcinosis, is also another challenge for health care professionals.

In this study, active ROM exercises were given for shoulder, spine, hip and knee joints of JDM patients. When the ROM values after the PA program were examined significant improvements were found in the right knee flexion, left knee flexion, spinal lateral flexion (right side) and spinal lateral flexion (left side) (p=0.007, p=0.046, p=0.039, respectively; Table 6). In the light of these findings, we may suggest that active ROM exercises for joints are a useful choice in the treatment of movement limitations and muscle contractures in patients with JDM.

In addition, the left shoulder flexion value of the JDM group was also increased after the PA program (p=0.024; Table 6). Depending on this finding, we may speculate that the dominant limb of the patients included in the study (n=22) was their right side and they were not using proximal muscles of the left shoulder as much as the right shoulder in their daily life. Therefore, the increase in ROM values in left shoulder may be due to increased use of left side by the ROM exercises. In the literature, medication and resting are recommended in the treatment of chronic rheumatic diseases. However, recent studies showed that exercise programs applied to JDM patients do not cause any adverse effects [23-25].

Helene Alexanderson et al. [24] applied resistance exercises (5 days/week during 12 weeks) within the home program of patients with JDM (n=11) and found no increase in the inflammation level and no negative process in JDM patients. As parallel to these findings we observed significant improvements in the CHAQ score after PA program in JDM patients of our study (p=0.000; Table 5). Although, regular PA program significantly improved their aerobic capacity the aerobic capacity of the JDM patients were found to be lower than their healthy counterparts significantly (Table 3 and Table 4). In addition to this, significant improvements in ROM levels and CHAQ scores reveal that PA programs can also improve the joint mobilization and quality of life in patients with JDM.

As the most of the patients included in our study lives in cities elsewhere than our medical center, the PA program we conducted in our study was not under supervision and was as homework. This situation may be considered as a limitation in our study. However, patients and their families are regularly called and reminded of the

exercises and their compliance with the program was questioned. In addition, regular walking and ROM exercises, which are preferred as activity models, do not require an economic cost for families. Also the intensity and the duration of the exercises in our PA program did not cause any complications in the clinical prognosis of our patients. For all these reasons, we think that all patients included in the study completed the PA program.

In conclusion, we believe that regular walking combined with active ROM exercises will be safe and beneficial in the management JDM treatment. Nevertheless, in the process of planning the PA program we also want to emphasize the importance of paying attention to 1) the involvement of the joint and/or muscle group, 2) the cardiopulmonary capacity of the patient. All parents of subjects gave informed consent to participate in the study, which was approved by the local ethics committees at Istanbul University. The study followed the World Medical Association's 2013 Declaration of Helsinki.

Conflict of Interest

The authors declare they have no conflicts of interest.

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