



The effects of hydrotherapy on muscle strength, body composition, and quality of life in boys with Duchenne dystrophy

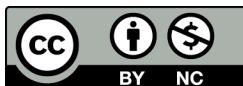
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Article Info	Abstract
<p>Original Article</p> <p>Article history: Received: 19 August 2021 Revised: 25 August 2021 Accepted: 04 October 2021 Published online: 05 December 2021</p> <p>Keywords: body composition, Duchenne muscular dystrophy, exercise, hydrotherapy, strength.</p>	<p>Background: Duchenne muscular dystrophy (DMD) is one of the most rampant x-linked recessive lethal genetic disease with prevalence of approximately 1 out of 3500-5000 newborn boys worldwide. DMD is mutations-induced in gene encoding dystrophin that prevent the production of the muscle isoform of dystrophin (Dp427m).</p> <p>Aim: This study aims to evaluate the impact of hydrotherapy on health-related quality of life and body composition changes, as well as how it affects on skeletal muscle strength in boys with DMD.</p> <p>Materials and Methods: Eight boys, ranging from 6 to 12 years old with DMD were enrolled in this study. The patients were randomly assigned to either a hydrotherapy group as the intervention group (Group 1; n=4) or a non-exercise group as the control group (Group 2; n=4), to receive 12 sessions of a hydrotherapy program. The patients were assessed for changes in muscle strength in the lower limbs and back, left and right quadriceps, left and right-hand grip, LBM, SMM, and general mobility and balance, before and after hydrotherapy.</p> <p>Results: Significant improvements in general mobility and balance were observed, along with a significant increase in muscle strength and lean body mass ($P<0.05$).</p> <p>Conclusion: This study suggests that hydrotherapy with a precise protocol of low to moderate intensity can improve general mobility and balance, increase muscle strength in the lower limbs and back, left and right quadriceps, left and right-hand grip, and LBM in patients with DMD. The increase in strength is accompanied by increase in LBM, which not only has implications for function but also has much broader impacts on the health-related quality of life in patients.</p>

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1. Introduction

Duchenne muscular dystrophy (DMD) is one of the most rampant x-linked recessive lethal genetic disease [1, 2], with prevalence of approximately 1 out of 3500-5000 newborn boys worldwide [3]. DMD is mutations-induced in gene encoding dystrophin that prevent the production of the muscle isoform of dystrophin (Dp427m) [4]. Thus, the absence or inactive of dystrophin protein leads to the replacement of muscle fibres by fat and connective tissue [5], the decrease of sarcolemma integrity [6], impaired mechanism of muscle regeneration and muscle injury [7]. Why does these happen occurs? Dystrophin play a fundamental role in stabilize and protect the muscle fiber during muscle contraction [8]. The initial symptoms appear with progressive muscle weakness in the lower limbs and frequent falls [9, 10] then the progression of the disease continue with more loss of muscle strength and stability, obesity, decreased ambulatory capacity, decreased upper limb function and eventually fatal cardiac and respiratory complications [11]. In fact, the pre-disposition of impaired muscular, respiratory and cardiac systems is immobility that resulting in considerable changes body composition in Patients with DMD, which highly affects on the health-related quality of life in patients [12].

Unfortunately, despite new and promising drugs and genetically therapeutic approaches (as if gene therapy has been limited to animal models) [13], there are no definitive and uncomplicated treatment [14]. Although, the drugs like Corticosteroids delay the loss of ambulation [15], but unfortunate effects are osteoporosis, cushing's syndrome, hypertension, hyperglycemia and short stature that hampers the use of

corticosteroids [16]. Given that there is a great demand for non-invasive treatments for DMD, thus hydrotherapy is one of the most cost-effective, non-invasive and feasible treatment's strategy for DMD [17]. The efficacy of various regular physical activity that is essential to maintain health, functioning and quality of life, have evaluated in numerous studies [18, 19].

One of the intriguing exercise facts that if properly prescribed and performed (the type, intensity, duration, and frequency) [20, 21] improve physiological capacities of muscle such as strength and endurance, with the adaptations dependent on the specific physiological [22]. Betterment in strength, will have a significant effect on the ability to perform activities of daily life [13, 23]. If increase of strength be accompanying by increase of muscle mass, such changes in body composition can also have metabolic benefits [24], and not only their implications on function, but also the much broader impacts on health and wellbeing [25]. This conclusion is based on highly abundant of evidence showing that strength training improves muscular strength and body composition [24].

Numerous studies have demonstrated that strength training can improve physical function [13]. The physiological results of resistance training like increased skeletal muscle mass, body composition change and metabolic changes to provide for increased strength and less fatigue during physical activity is obvious [13, 26].

On the base of available data, regular low-intensity exercise can maintain muscular strength and prevent muscle atrophy [21, 27], finally functional status and quality of life improve in patients with DMD [28, 29]. From a quality-of-life perspective, these are very desirable outcomes for individuals with DMD, but on

the other hand, there is the major limitation to exercise in patients with DMD. This limit is intensive eccentric muscle exercise that could exacerbate muscle damage and cause the increased connective tissue deposition [21, 27]. How can we best perform the exercise prescription without injury that might result from the physical activity in patients with DMD?

Currently, prescription modality which be safe and make exercise feasible for patients and can use in all phases of disease progression for DMD patients is aqua therapy (perform exercise in water) [30, 31]. Hydrotherapy is including non-weight physical activities in water [32], in order to keep DMD patients physically active [33, 35], which does not put extra pressure on muscles the buoyant forces of water are cause to reduce joint loading and facilitate antigravity movements, resistive properties promote postural and extremity strengthening, transfer in water when compared with air may reduce tone and non-volitional movements [33, 34]. Also allows DMD patients to improve their balance and gait in without risk of falling or pain [31, 35], but if they perform such movements out of the water, they could experience significant pain or problems in physical functioning [31].

There are many clinical studies conducted with DMD patients; however, there is a lack of evidence and researches investigating the therapeutic effects of exercise like hydrotherapy on physiological indexes in individuals with DMD [33]. In this respect, this study fills a gap in the literature and provides a standpoint for future research. Consequently, the aims of this study are evaluating the impact of hydrotherapy on health-related quality of life and body composition changes in boys with DMD, also how hydrotherapy affects

the strength of the skeletal muscle in DMD patients.

2. Methods

2.1. Subjects

This was a fundamental, quasi-experimental qualitative study that assessment the impact of hydrotherapy on health-related quality of life, the strength of the skeletal muscle body, and composition changes in boys with DMD. The statistical population consisted of boys with DMD who were members of the Iranian Dystrophy Association. Eight of them aged 6–12 years were selected as the sample using purposive and convenience sampling to participated in this study.

The method of calculating the sample size was considering G power software. The inclusion criteria for this study were male subjects with Duchenne dystrophy and no mental health issues. To control the effect of the circadian cycle, the test was performed in the morning between 8 and 11. The variables measured in this study changes body composition such as Lean Body Mass (LBM) and Skeletal Muscle Mass (SMM), health-related quality of life (General mobility and balance), strength of the skeletal muscle (Lower limbs and back, Quadriceps, Hand grip).

It was mentioned to the subjects that, if they did not want to cooperate for any reason in any time of the study, they could freely withdraw from the study. The participants were invited to the Human Physiology Laboratory, Faculty of Sport Sciences and wellness, University of Tehran, for the initial evaluations and the pretest. Each participant was given the health and medical records forms, the informed consent form and the certificate of participation.

All patients were randomly assigned to

two groups, including a hydrotherapy group as the intervention group (Group 1; n=4) and a non-exercise group as the control group (Group 2; n=4). Both groups underwent the same 5 physical function tests at similar time intervals. The strength of the upper and lower body and body composition were measured in DMD patients. Hydrotherapy intervention in Group 1, the patients performed the hydrotherapy under one-by-one supervision of a hydro therapist. This hydro therapist had 8 to 10 years of experience in providing pediatric hydro therapy intervention. The hydrotherapy program consisted of 12 sessions (3 times per week for 4 weeks) in a swimming pool at 32°C. Each session lasted 45 min. Each of 4 patients performed the same exercises; however, the exercises were individualized for each participant based on the number of repetitions and the intensity level which the patients were capable of.

2.2. Tests (*pre-test-post-test*)

The participants were invited to the Human Physiology Laboratory, Faculty of Physical Education and Sport Sciences, University of Tehran, for the initial evaluations and the pretest. An informed consent was obtained from each participant and each parent before the examination. Then, their height and weight were evaluated by scale and height meter and their combination was determined by Body compound analyzer (body composition evaluation to describe percentage, LBM and, SMM). These percentages are of most interest, because it can be very helpful in assessing health [36, 37]. Then, the tester, patients' strength of quadriceps femoris muscle performed MMT (assessment of the maximum force a muscle is capable of generating, and determining the quality of eccentric muscle

strength) according to the test of quadriceps femoris muscle [38].

A handheld device recorded the temporal course of the applied force. In the lowest degrees, only movements of tendons or muscles are observed. But, in the upper degrees, the focus lies on the ability to hold against an external pressure applied by the examiner [39]. Also, the mean of three isometric contractions was recorded for each participant.

The strength of the upper and lower body was also measured in the same way, using a Back-leg-chest Dynamometer (measurement and evaluation of muscle strength and endurance of lower limbs and back isometrically) [40, 41]. Then, Handgrip Strength Test is a measure of muscular strength or the maximum force/tension generated by one's forearm muscles, and it can be used as a screening tool for the measurement of upper body strength and overall strength [42]. It is most useful when multiple measurements are taken over time to track performance [43].

Finally, the Timed Up and Go Test (TUG) was performed to measure general mobility and balance (those in a wheelchair could not do this test). In the timed up and go (TUG) test, subjects were asked to rise from a standard armchair, walk to a marker 3m away, turn, walk back, and sit down again [44, 45]. The test is a reliable and valid test for quantifying functional mobility that may also be useful in following clinical change over time. It is a simple screening test that is a sensitive and specific measure of probability for falls among DMD patient [46].

A recent study found that the TUG test is a strong mortality predictor [47]. Those in the control group were not asked to attend any sports and functional activities or physiotherapy, occupational therapy, or cell

therapy intervention during the study (4 weeks). Twenty-four hours after the last hydrotherapy session, the participants repeated, once again, all laboratory tests as in the pretest.

2.3. Hydrotherapy protocol

The intervention group only participated in the hydrotherapy program for four weeks and did not receive any other sports, functional, physiotherapy, or occupational therapy interventions before or during the study. This intervention was performed in twelve 45-min sessions 4 times a week under the supervision of an experienced hydro therapist. In each session, the hydro therapist guided and helped each participant, who had a companion, to perform the protocol. For a few days, the hydro therapist tried to gradually introduce the participants with water because most of them had never entered a pool before. The tools required for the field test included a private hydrotherapy pool equipped for the disabled with an aqua temperature suitable for the patient (32°C) and a ramp for entering the aqua and proper ventilation for the patients [48]. All kinds of hydrotherapy equipment, ranging from light to heavy inflatable balls in different sizes, thera bands with different resistances, special loop band for feet and hand, aquatic noodles, life jackets, aqua dumbbells, hand paddle, and paddle boards [49]. Table 1 provides more details on how to perform the hydrotherapy protocol.

2.4. Statistical analysis

The data on the research variables were analyzed using descriptive statistics. In addition, the independent samples t-test and the dependent samples t-test were employed for the inter-group and intra-group comparison of the dependent variables. The significance level in all statistical tests was

considered to be $\alpha=0.05$. All statistical calculations were done by SPSS21 statistical software.

3. Results

The results of the independent t-test of the pre-test and post-test for the control and intervention groups are presented separately in Table 2.

The results of the independent t-test showed that there is a significant difference between the percentage of changes in lower limbs and back strength, left quadriceps strength, right quadriceps strength, left hand grip strength, and right-hand grip strength of patients with DMD, in the control and experimental groups (respectively, [T=3.13, $P=0.020$], [T=15.38, $P=0.001$], [T=13.91, $P=0.001$], [T=5.55, $P=0.003$], [T=9.38, $P=0.002$]). Therefore, it could be concluded that hydrotherapy had an effect on the strength of the back, quadriceps and hand grip of patients with Duchenne dystrophy and has increased muscle strength (Table 2).

The results of the independent t-test showed that there is no a significant difference between the percentage of changes in SMM of patients with DMD, in the control and experimental groups [T=-0.99, $P=0.059$]. Therefore, it can be concluded that hydrotherapy had not an effect on the SMM of patients with Duchenne dystrophy (Table 2).

The results of the independent t-test showed that there is a significant difference between the percentage of changes in LBM ([T=4.318, $P=0.005$]) and General mobility and balance ([T=-5.93, $P=0.006$]) of patients with DMD, in the control and experimental groups. Therefore, it can be concluded that hydrotherapy has had an effect on LBM, general mobility and balance of patients with DMD (Table 2).

The effects of hydrotherapy on boys with Duchenne dystrophy

Table 1. Hydrotherapy protocol

Target	Tools	Frequency	Duration (min)	Intensity**	Reps	Set	Rest between sets ***	Overload	Type
Warm-up	-	3	10	low	8	1	-	1	General warm up and Joint mobility: Marching in pool while swinging their arms/ arm circles and shoulder shrugs/ swinging toe touches/ leg swings (forward)/ leg swings (side to side)/ hip rotations (like stepping over a fence)/ hip circles
Isometric and Isotonic resistance training	Thera bands with different resistances, special loop band for feet and hand Water dumbbells	3	15	Low-moderate	6	2	1 min	2	Isometric: Lateral raise/ chest press/ leg press/ seated calf press/ triceps press (with thera band and loop/ hold each end of it 8 Second) Isotonic: Bicep curls/ flies/ karate punch/ back wall glide/ high-knee lift extensions (with aqua dumbbells)
Breathing exercises	Ranging from light to heavy inflatable balls, small balloon	3	5	Low-moderate	10	2	30 sec	2	Pursed-Lips breathing and blowing out balls on water/ making a gurgling sound (like when water boils or flows) under water/ inflate a balloon
Aerobic exercise	Hand paddle, and paddle boards	3	10	low	8	2	1min	2	Leg kicks/hand kicks
Cool-down	-	1	5	low	2	1	20s	1	PNF technique (contract-relax-antagonist-contract method) *for quadratus femoris, hamstring muscles, biceps, triceps, hip flexors and back muscles.

*Contract-relax-antagonist-contract method (CCRAC) method: 1. target muscle (TM) being lengthened (“stretched”); 2. hold in position while the person contracts (activates) the agonist (TM) to about its 50-60% of maximum isometric contraction for 4-6 sec; 3. straight away then activate the antagonist muscle for 4-6 seconds, defined as the muscle that opposes the action of another; “the biceps and triceps are antagonistic muscles; 4. the muscles are then relaxed for 20 sec before performing another PNF technique [50]

**Methods of measuring intensity is perceived exertion (Borg rating of perceived exertion scale)

***Rest between set in addition to the time mentioned is based on the fatigue and needs of the person

Table 2. The pretest and posttest descriptive data for the control and intervention groups

Variable	Groups	Pre	Post	P-value
Lower limbs and back strength	1	6.00±3.46	12.07±5.30	0.020*
	2	8.25±4.71	8.50±3.10	
The quadriceps strength (left leg)	1	3.12±0.79	6.62±1.31	0.001*
	2	5.92±2.49	2.72±1.28	
The quadriceps strength (right leg)	1	2.62±0.29	5.30±1.28	0.001*
	2	5.97±3.26	2.95±1.57	
Hand grip strength (left hand)	1	2.77±1.24	6.62±1.84	0.003*
	2	3.07±0.54	1.17±0.09	
Hand grip strength (right hand)	1	3.02±1.60	6.92±2.69	0.002*
	2	3.05±0.38	1.35±0.20	
SMM	1	11.72±6.87	19.02±0.41	0.059
	2	12.15±2.63	6.70±4.31	
LBM	1	19.85±1.62	25.75±2.30	0.005*
	2	15.15±4.72	13.47±2.45	
General mobility and Balance	1	17.55±5.07	13.25±4.54	0.006*
	2	11.12±7.24	14.99±8.10	

*P<0.05

4. Discussion

This study has conducted to investigate the effects of hydrotherapy on health-related quality of life and changes in body composition and skeletal muscle strength in DMD patients.

Due to the degenerative nature of this disease, the strength of the skeletal muscles decreases, as a result, the person with DMD becomes immobile over time to the point where he is not even able to perform his daily activities, and also makes them face disorders of the muscular, cardiac and respiratory systems. This greatly affects their quality of life and brings their death forward.

This conclusion is based on highly abundant of evidence and studies showing that strength training improves muscular strength [24], and increasing muscle strength can affect cardiorespiratory function, flexibility, and increase range of motion, balance, and prevention of falling.

Available studies indicate that exercise

improve physiological capacities of muscle such as strength and endurance [22]. Also, betterment in strength will have a significant effect on the ability to perform activities of daily life [13, 23]. Numerous studies evince that regular low or moderate intensity exercise can maintain muscular strength and prevent muscle atrophy [21, 27], finally improving functional status and quality of life in patients with DMD [28, 29]. According to the reported evidence and studies, although few, considering the physical limitations of DMD and the hydrostatic effects of water, performing exercises to improve strength in the aqua environment is a suitable and safe solution [51]. Given, any improvement in muscle strength and function or steady maintenance of function over time could be considered a positive outcome.

The results of our study are in line with the mentioned studies. Based on the first results of the present study, we conclude that people with DMD benefit from

hydrotherapy to increase muscle strength lower limbs and back, left and right quadriceps, left and right-hand grip.

The second findings from the current research are that hydrotherapy has improved the changes in body composition and increased LBM values, but statistically it had no effect on SMM. However, since the differences found in this study were not significant, this cannot be concluded with certainty. The lack of positive findings in SMM can be partially explained by the short duration of the program or the small sample size in this study.

Researchers reported in a study that if increase of strength be accompanying by increase of muscle mass, such changes in body composition can also have metabolic benefits [24], and not only their implications on function, but also the much broader impacts on health and wellbeing [25]. In patients with DMD the replacement of muscle fibers by fat and connective tissue occur [5]. Additionally, the pre-disposition of impaired muscular, respiratory and cardiac systems is immobility and obesity that resulting in considerable changes in body composition in patients with DMD, which highly affects on the health-related quality of life in patients [12].

Researchers have reported that strength training improves muscular strength and body composition [24]. Also, in a study, the physiological results of resistance training like increased LBM, body composition change, and metabolic changes to provide for increased strength and less fatigue during physical activity has characterized [13, 26].

The results of our study are in line with the mentioned studies and hydrotherapy cause increase LBM. Relationship between increase strength of muscles and LBW directly affect to decrease obesity. Muscles,

even at rest, require energy in the form of calories, while fat cells do not. So, the greater amount of lean muscle tissue, the more calories burn throughout the day, decreasing the likelihood of excessive fat accumulation. Eventually, the health-related quality of life in patients of DMD improves [52]. Based on the subsequent results of this study, we conclude that people with DMD benefit from hydrotherapy to improve general mobility and balance. The result in the present study is consistent with the past studies. Past studies have concluded that reduce of probability for falls among DMD patient through hydrotherapy, instead increase general mobility and balance.

5. Conclusions

Duchenne muscular dystrophy is being most common and severe genetic diseases. DMD lead to muscle weakness, loss of physical capacity, obesity, and eventually early death. There remains no effective and without side effects cure for DMD, and due to the advantages and characteristics of exercising in the aqua environment, which is a safe environment in terms of reducing the pressure on the body, the resistance of water is greater than air and strengthening. Hydrotherapy is one of the most cost-effective, non-invasive and feasible treatment's strategy for DMD [17]. Based on studies, although they were very few and indirect, we can conclude that hydrotherapy, with a precise protocol low to moderate intensity, cause improve general mobility and balance and increase muscle strength lower limbs and back, left and right quadriceps, left and right-hand grip, and LBM in patients with DMD. Also, increase of strength be accompanying by increase of LBM, not only had their implications on function, but also the much broader impacts on the health-related quality of life in

patients [25]. Hydrotherapy has shown a light of hope in the treatment and recovery of Duchenne dystrophy patients, which requires comprehensive studies by physiology researchers. It appears that future research should focus more on hydrotherapy in larger sample sizes.

Muscular dystrophy is one of the genetic diseases that has caused many health-medical and economic-social problems for human societies and has attracted the attention of many geneticists, neurologists and other specialists in various fields in recent decades [53]. Over the last few years, there has been considerable development of diagnosis and therapeutics for DMD, but current treatments do not cure the disease, therefore therapy is limited to the management of symptoms, delay the onset or slow down the progression [54] including drug (Corticosteroids) and gene interventions. Although, the drugs like Corticosteroids unfortunate side effects including osteoporosis, Cushing's syndrome, hypertension, hyperglycemia and, short stature hampers the use of corticosteroids [55, 56], as if gene therapy has been limited to animal models. It is very costly and time-consuming, and not everyone can afford this kind of treatment [57, 58, 59].

Given, there is a great demand for non-invasive treatments for DMD. Considering its prevalence and the very heavy financial and human costs and the loss of life expectancy of the sufferers, this incurable disease should be managed and action should be taken towards the optimal and effective treatment of dystrophy. Mainly, hydrotherapy are one of the most cost-effective and non-invasive treatments strategy for DMD [18, 23], even is as an adjunct therapy in prevention of complications, arrest or slowing of

progression of the pathophysiology, improvement in the quality of life with better adjustment [13, 60]. In the same direction in patients with DMD, there have been studies conducted on pathophysiology mechanisms of the disease but have been not studies on the adaptation to physical activity [13, 61]. In this respect, this study fills a gap in the literature and provides a standpoint for future research.

Conflict of interest

The authors declared no conflicts of interest.

Authors' contributions

All authors contributed to the original idea, study design.

Ethical considerations

The author has completely considered ethical issues, including informed consent, plagiarism, data fabrication, misconduct, and/or falsification, double publication and/or redundancy, submission, etc.

Data availability

The dataset generated and analyzed during the current study is available from the corresponding author on reasonable request.

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