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ORIGINAL ARTICLE

Comparison of telerehabilitation versus home-based video exercise in patients with Duchenne muscular dystrophy: a single-blind randomized study

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Abstract

Introduction Patients with Duchenne muscular dystrophy (DMD) have lost their access to on-site rehabilitation due to the COVID-19 pandemic. Telerehabilitation can be a viable approach for these patients to protect their muscle strength and functional status. The aim of this study is to compare telerehabilitation with home-based video exercises.

Patients and methods Male, ambulatory DMD patients were randomized into telerehabilitation and video-exercise groups. Nineteen patients were included in the final analyses. Telerehabilitation consisted of live online exercises, while video exercise implemented a pre-recorded video as a home-based program. Both programs spanned 8 weeks, three times a week. Patients' muscle strength with a hand-held dynamometer, Quick Motor Function Test, North-Star Ambulatory Assessment (NSAA), 6-Minute Walk Test (6MWT) and Caregiver Burden were recorded before and after treatment.

Results The 6MWT of the telerehabilitation group was391.26 ± 95.08 m before and387.75 ± 210.93 after treatment ($p = 0.94$) and 327.46 ± 103.88 m before treatment and313.77 ± 114.55 after treatment in video group ($p = 0.63$). The mean NSAA score of the telerehabilitation group were26.70 ± 8.04 before treatment and 25.20 ± 11.33 after treatment ($p = 0.24$). In the video group scores were 21.66 ± 6.65 before to 22.00 ± 8.61 after treatment ($p = 0.87$). There were no significant changes between groups at the end of the treatments. The telerehabilitation group's neck extension, bilateral shoulder abduction, and left shoulder flexion, bilateral knee flexion and extension, bilateral ankle dorsiflexion, and left ankle plantar flexion strength improved significantly and were better than the video group ($p < 0.05$ for all measurements).

Conclusion A telerehabilitation approach is superior in improving muscle strength than a video-based home exercise, but none of the programs improved functional outcomes in ambulatory patients with DMD.

Introduction

Duchenne muscular dystrophy (DMD), a recessive transition disorder associated with the X chromosome, is a common childhood condition that affects males almost exclusively. It is the most common muscular dystrophy with an incidence of 1/5000 boys and is characterized by muscle weakness in early childhood [1]. DMD is a genetic condition in which dystrophin deficiency leads to an imbalance of the glycoprotein complex of muscle sarcoma. The glycoprotein complex is necessary to maintain regular muscle activity [2]. Continuous degeneration of muscle fibers in DMD patients leads to progressive loss of functional ability and decreases the strength of skeletal, respiratory muscles, myocardium [3–5]. In DMD, symptoms are usually evident between 3–5 years of age. These symptoms include walking disorders, difficulty rising from the ground, and frequent falls. Weakness is more prominent in proximal than distal muscles and the lower limb more than the upper limb [6].

Comprehensive and prospective preventive rehabilitation management in DMD focuses on protecting and maintaining optimum muscle strength, minimizing the progression of weakness as much as possible, preventing progressive contracture and deformity, supporting optimal cardiorespiratory function, providing adaptive equipment, and assistive

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technology. The aims of rehabilitation can be summarized as improving participation in school, family, and social life and optimizing the quality of life. Prevention of contracture and deformity requires daily active assistive and passive stretching of the soft tissues and muscles of the joints that have a feeling of tension. Moreover, it should not be overlooked that muscle strengthening is also important, but it is known that high-intensity exercises increase muscle damage and fibrosis. Low-intensity or moderate-intensity aerobic exercise may improve skeletal muscle function, reduce cardiac decline, and increase respiratory capacity [7].

The COVID-19 pandemic has affected all aspects of healthcare delivery, making it difficult for patients to receive institutional rehabilitation services, and admissions to many clinics have been stopped or restricted. Existing literature suggests that the implementation of telerehabilitation services can provide adequate care for children and adolescents with special needs during the COVID-19 process [8]. Telerehabilitation is the remote delivery of rehabilitation services using information and communication technologies [9]. In systematic reviews that included studies examining the effectiveness of telerehabilitation in patients with musculoskeletal problems, it was reported that telerehabilitation was as effective as face-to-face methods on pain, functional outcomes, and other outcome measures [9].

In a study conducted to investigate the rehabilitation status of patients with DMD in the pandemic, to create an online rehabilitation program and motor assessment, and to determine the telerehabilitation needs in this group, online studies were conducted for 69 DMD patients together with their families; evaluations and treatments were carried out online. It has been suggested that online videos can be an acceptable alternative to caregivers of DMD patients than live workshops [10]. However, there is not a previous study that compared the effectiveness of these two remote approaches, telerehabilitation and video-based home exercise program, in patients with DMD.

Considering the patients with DMD who could not receive institutional rehabilitation service during the COVID-19 pandemic period, the aim of this study is to maintain their rehabilitation service through telerehabilitation that includes live video sessions with a physiotherapist or a pre-recorded video based home exercise program and, to compare these two approaches in two groups on their effects on motor function, functional capacity, muscle strength, and caregiver burden.

Methods

The study was carried out at a tertiary university hospital that also is a center for DMD patient care between February 2021 and May 2021. The study was approved by the local ethics committee. The ethical approval number is 2021–353. This study had been registered to clinicaltirals.gov with the registration number of 'NCT04782440'. Before the study, all participants were given detailed information about the study. Both the parents and the patients gave written and verbal informed consent.

Male patients between the ages of 6–15 who were followed up with the diagnosis of DMD, did not lose their ability to ambulate, did not have mental retardation or cognitive impairment that prevented the tests from being used in the evaluation, were included in the study. Patients diagnosed with DMD who lost their ability to ambulate had undergone spinal or orthopedic surgery in the last 6 months and had serious comorbidities other than neuromuscular disease were excluded from the study. All patients that were invited to the study were being followed up by the Pediatric Neurology clinic and were contacted by the doctors who were involved in their care for recruitment. The study was explained to the patients in detail regarding their potential participation in a telerehabilitation or a video based home exercise program. The evaluations were explained to the patients before recruitment. The recruiters were blind to the patient allocation procedure and were not involved in the evaluation processes. Patients were randomized into a telerehabilitation program (telerehabilitation group)or video-based home exercise program (video exercise group) by a computer-generated list. From a total population of 25 suitable patients with DMD, 19 patients were included in the final analyses. 2 patients were not able to ambulate properly, one patient declined to participate, so they were excluded at the beginning of the study. During the study, three patients in the video exercise group could not participate in the second assessment. Therefore, they were not included in the final analyses. The patient flowchart can be seen in Fig. 1.

Exercise program

The telerehabilitation group participated in a one-on-one exercise program under the supervision of a physiotherapist for 30–40 min, 3 days a week for 8 weeks. All patients and the physiotherapists used laptops during the telerehabilitation sessions. The physiotherapist gave necessary feedback and encouragement during the sessions since they were live and allowed interaction between the physiotherapist and the patient. The exercises were given to the Video Exercise Group as recorded videos given via a streaming video, and the patients were asked to perform these exercises once a day for 8 weeks, 3 days a week at home. They did not receive any feedback during the study from the physiotherapists and were just encouraged to do the exercises on a regular basis. The content of the exercise program was the same for both groups (provided as a supplementary file). The exercises were determined in such a way that they can be done at

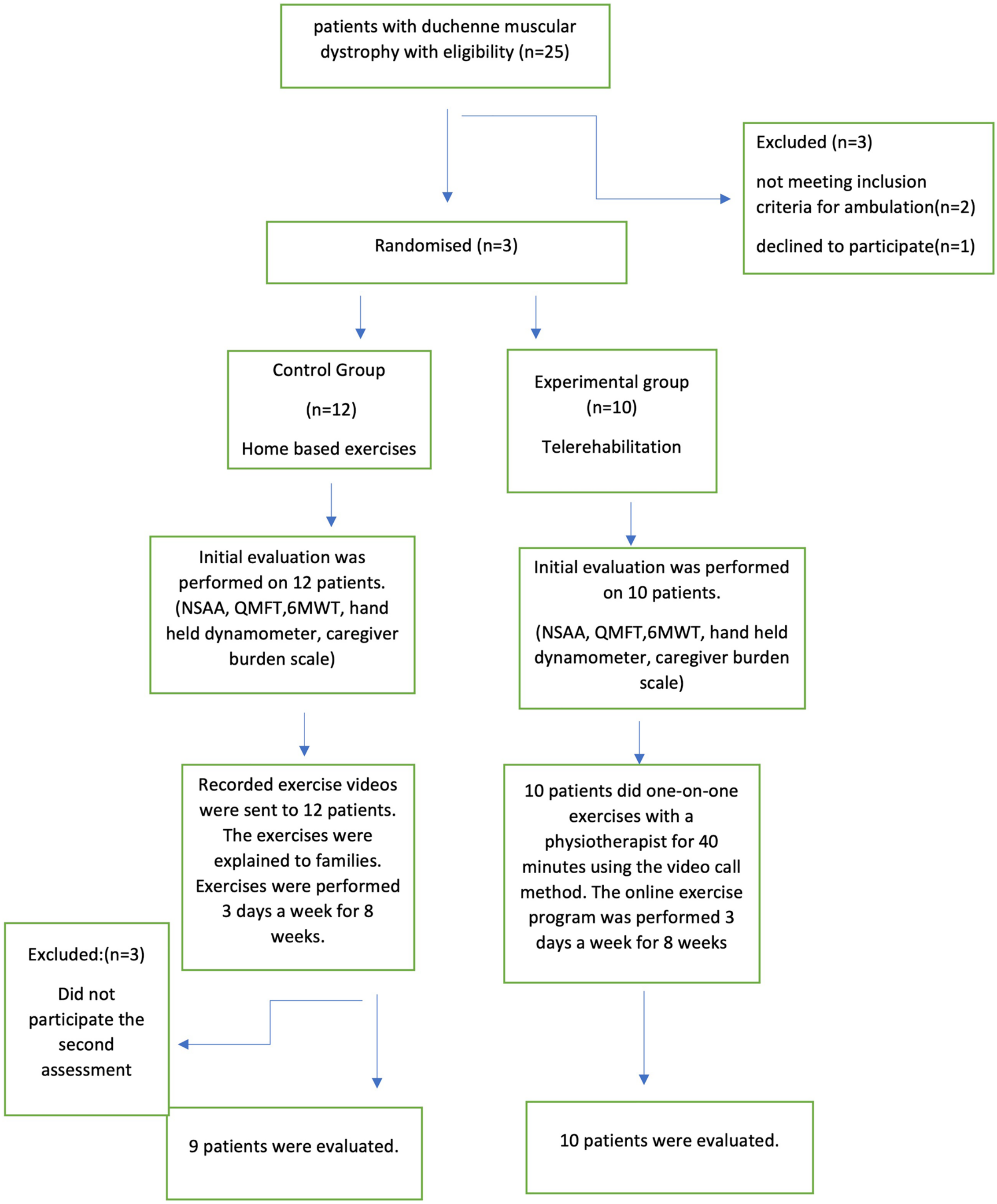

Fig. 1 Patient flowchart

home, and no equipment is needed. This exercise program consisted of low-moderate intensity aerobic exercise that includes rhythmic normal joint movements that can be done sitting in a chair, chin tuck exercise, quadriceps strengthening exercise in sitting position (using body weight against gravity), posterior pelvic tilt exercise, gluteus maximus isometric strengthening exercise, bridge exercise, hip lying on the side exercise to strengthen the abductors (using body weight against gravity) included stretching exercises for the pectoral muscles, trunk lateral flexors, iliotibial band, hamstring, and gastro-soleus muscles (with the help of a towel or sheet). In the Telerehabilitation Group, each exercise was performed with 10 repetitions, and patients were given rest periods between exercises. The Video Exercise Group was likewise asked to do each exercise for 10 repetitions and to leave rest periods between exercises.

Patients' participation was recorded by the therapist that implemented the session in the telerehabilitation group and by the parents in the video exercise group.

Evaluations

All assessments were done face-to-face in a clinical setting by a single researcher who is blind to the allocation of the patient, who was not a part of any therapy session and who were not involved in patient recruitment process. Demographic information such as age, height, weight, body mass index (BMI), and cumulative corticosteroid doses were recorded in the cases included in the study. Patients' weight and height measurements were done with a DR-MOD.85 model scale (Baskul Ticaret, Istanbul, Turkey). Patients' corticosteroid doses were calculated based on the fact that the patients received 0.35–0.40 mg/kg/g prednisolone multiplied by the total time of use and patients' weight in kilograms. Quick Motor Function Test (QMFT), North Star Ambulation Evaluation Ambulatory Assessment (NSAA), muscle strength test with Hand Held Dynamometer (Baseline Push–Pull Dynamometer, Fabrication Enterprises, New York, USA), 6 Minute Walk Test (6MWT), and Caregiver Burden Scale were used to evaluate the findings in both groups two times: at the beginning and end of the exercise program.

Functional assessments

QMFT, which is used to evaluate motor function, is a test that does not require special equipment and can be performed by a doctor or physiotherapist in a clinical setting. The test consists of 16 items and takes approximately 15 min. The evaluator observes the patient's performance, and if the items can be performed on both the left and right extremities, the right side is considered. The items of each item are scored between 0 and 4 points, and the total score varies between 0 and 64. Higher scores indicate better motor function [11].

The North Star Ambulatory Assessment was used to evaluate the ambulation. It is a scale specific to patients with ambulatory DMD and consists of 17 items. Scale items are scored between 0 and 2. The total score ranges from 0 to 34. All items are tested without trunk brace or leg orthoses and are usually completed in 15 min [12].

The 6 Minute Walk Test was used to evaluate the functional capacities of the patients. The test is calculated by measuring the distance walked along the 30-m-long track, as recommended in the American Thoracic Society manual [13]. The patients were given detailed information about the test, and the patient was asked to walk the longest distance he could walk in 6 min on the 30-m track until he heard the command " over ". It was explained that if they felt too much shortness of breath and fatigue while walking, they could either rest or terminate the test. 6 MWT were measured in meters.

Strength measurement

Hand-held dynamometer was used to evaluate muscle strength (baseline digital push–pull dynamometer). During the evaluation, an isometric contraction was requested from the patient as much as possible, and the applied pressure is gradually increased for about 3 s. The applied pressure was stopped at the point where the patient cannot bear it [14–16]. Each muscle was tested three times with 10-s intervals between measurements. Particular attention was paid to the anatomical region where the measuring device had been precisely placed [17]. The mean intra-rater correlation coefficient of the measurements was 0.89 showing good agreement for each measurement.

Caregiver burden

The Zarit Caregiver Burden Scale was applied to the families of patients with DMD at the beginning and end of the study. It is a scale used to evaluate the stress experienced by caregivers of individuals in need of care. The scale, which can be filled by the caregivers themselves or by the researcher, consists of 22 statements that determine the effect of caregiving on the individual's life. The scale has a Likert-type rating ranging from 0 to 4 as never, rarely, sometimes, often, or almost always. A minimum of 0 and a maximum of 88 points can be obtained from the scale. A high score on the scale indicates a high level of distress [18].

Statistical analyses

Sampling size calculations were done by using G power. There is no study in the literature that previously measured the effect of internet-based exercises performed by DMD patients and their effects on proximal muscle strength. Considering the relatively rare nature of the disease and the highly selective inclusion criteria, a posthoc analysis was made. This analysis showed that an effect size of nine patients in each group was sufficient to reach an effect size of 0.9 with a power of 80% for the change in deltoid muscle power. All data analyses were done by another researcher who were not blind to the allocation of the patients and the research had been unblinded at this stage, since the database clearly showed which patient was allocated to which group. The distribution of the data was analyzed with Shapiro-Wilks's test, and it was found to be normal. Basic descriptive analyses were used for the calculation of frequencies, means, and standard deviations. For baseline differences between groups, independent samples *t* test were used. Two-way mixed ANOVA was used for within-group and between-group analyses. A p value below 0.05 was considered significant since there were only baseline and after-treatment measurements. All statistical analyses were done by SPSS version 20.0.

Results

There were no significant differences between the ages, functional status, caregiver burden score, and cumulative steroid doses between each group. Patients in the telerehabilitation group had better strength in their shoulder abductors, right hip flexors, and right hip abductors at their baseline measurements (Table 1).

The mean 6-min walk test of the telerehabilitation group was 391.26 ± 95.08 m before treatment and it was 387.75 ± 210.93 after treatment, showing no significant differences ($p = 0.94$). Similarly, these values were 327.46 ± 103.88 m before treatment and 313.77 ± 114.55 after treatment in the video exercise group, again without changing significantly ($p = 0.63$). The mean NSAA score of the telerehabilitation group were 26.70 ± 8.04 before treatment and 25.20 ± 11.33 after treatment, showing no significant changes ($p = 0.24$). Similarly, the video exercise group scores did not change significantly, from 21.66 ± 6.65 before treatment to 22.00 ± 8.61 after treatment ($p = 0.87$). There

Table 1 Demographic characteristics and average muscle strength of the patients at the baseline

	Telerehabilitation group Mean (SD)	Video exercise group Mean (SD)	*p* value
Age (years)	8.80 (2.93)	7.00 (2.00)	0.14
BMI (kg/m^2)	16.90 (2.56)	15.80 (3.25)	0.75
6MWT (m)	391.26 (95.08)	327.46(103.88)	0.18
NSAA score	26.70 (8.04)	21.66 (6.65)	0.16
QMFT score	52.40 (13.60)	45.66 (11.78)	0.27
Caregiver burden score	29.70 (15.05)	27.12 (19.69)	0.76
Cumulative prednisolone dose (g)	3.69 (5.12)	3.12 (6.18)	0.81
Neck flexion (kg)	1.69 (0.87)	1.03 (0.5)	0.07
Neck extension (kg)	2.28 (0.90)	1.87 (0.66)	0.28
R shoulder abduction (kg)	2.82 (0.74)	1.98 (0.53)	0.01*
L shoulder abduction (kg)	2.52 (0.80)	1.76 (0.62)	0.04*
R elbow flexion (kg)	2.75 (1.38)	1.76 (0.52)	0.07
L elbow flexion (kg)	2.78 (1.59)	1.84 (0.78)	0.13
R elbow extension (kg)	2.59 (1.15)	1.62 (0.53)	0.03*
L elbow extension (kg)	2.55 (1.04)	1.77 (0.66)	0.08
R hip flexion (kg)	3.25 (1.24)	1.95 (0.80)	0.02*
L hip flexion (kg)	3.10 (1.14)	2.27 (0.98)	0.11
R hip abduction (kg)	3.62 (1.32)	2.40 (0.91)	0.04*
L hip abduction (kg)	3.10 (1.14)	2.27 (0.98)	0.09
R knee flexion (kg)	3.14 (1.08)	2.27 (0.78)	0.07
L knee flexion (kg)	2.94 (1.09)	2.28 (0.92)	0.18
R knee extension (kg)	2.98 (1.11)	2.65 (1.15)	0.54
L knee extension (kg)	3.09 (1.00)	2.48 (1.01)	0.23
R wrist flexion (kg)	1.75 (0.69)	1.17 (0.36)	0.41
L wrist flexion (kg)	1.75 (0.84)	1.28 (0.42)	0.15
R wrist extension (kg)	1.82 (0.49)	1.25 (0.43)	0.17
L wrist extension (kg)	1.84 (0.66)	1.11 (0.33)	0.09
R ankle dorsiflexion (kg)	2.71 (0.86)	1.97 (0.82)	0.07
L ankle dorsiflexion (kg)	2.58 (0.52)	2.20 (0.95)	0.36
R ankle plantarflexion (kg)	3.12 (1.14)	2.58 (0.80)	0.26
L ankle plantarflexion (kg)	2.99 (1.63)	2.20 (0.76)	0.20

BMI body mass index, *6MWT* 6 min walking test, *NSAA* north star ambulatory assessment, *QMFT* quick motor function test, *R* right, *L* left, *SD* standard deviation

*Marks $p < 0.05$

were no significant changes between groups at the end of the treatments either, showing no change in functional outcomes. There were no significant differences in either group in QMFT and Caregiver burden scores. These findings are summarized in Table 2. The telerehabilitation group's neck extension, bilateral shoulder abduction, and left shoulder flexion strength improved significantly and were significantly better than the video exercise group after treatment (Table 3). Similarly, bilateral knee flexion and extension, bilateral ankle dorsiflexion, and left ankle plantar flexion strength improved significantly and were better in the telerehabilitation group when compared to the video group in the lower extremities (Table 4). There was no significant improvement in any of the parameters measured in the video exercise group.

There were no adverse events directly related to rehabilitation protocol or video exercises. Since each patient completed a total number of 24 sessions, the total number of sessions was calculated as 240 in the telerehabilitation group and 216 in the video exercise group. The total number of participation in each of the sessions were 234/240 (97%) in the telerehabilitation group when compared to 180/216 (83.3%) in the home-based exercise group.

Discussion

This single-blind randomized controlled study demonstrated that a telerehabilitation approach was superior to improving muscle strength when compared to a video exercise program. But the improvements in muscle strength did not manifest as functional improvement and did not affect caregiver burden levels.

Table 2 Mean differences of 6-min walk test, North Star Ambulation Assessment, Quick Motor Function Test and caregiver burden within and between groups

	Group	Baseline Mean (SD)	End of treatment Mean (SD)	Mean difference within group (95% CI of the difference) *p* value
Six-Minute Walk Test (m)	Telerehabilitation group	391.26 (95.08)	387.75 (210.93)	3.51 (−98.54 to 105.56) $p=0.94$
	Video exercise group	327.46 (103.88)	313.77 (114.55)	13.68 (−49.28 to −76.66) $p=0.63$
	Mean difference between groups (95% CI of the difference) *p* value	63.79 (−32.49 to 160.07) $p=0.18$	73.97 (−93.17 to 241.12) $p=0.36$	
North Star Ambulation Assessment (out of 34)	Telerehabilitation group	26.70 (8.04)	25.20 (11.33)	1.50 (−1.22 to −4.22) $p=0.24$
	Video exercise group	21.66 (6.65)	22.00 (8.61)	−0.33 (−4.79 to −4.13) $p=0.87$
	Mean difference between groups (95% CI of the difference) *p* value	5.03 (−2.16 to −12.22) $p=0.16$	3.20 (−6.63 to 13.03) $p=0.50$	
Quick Motor Function Test (out of 64)	Telerehabilitation group	52.40 (13.60)	50.60 (17.72)	1.80 (−2.12 to 5.72) $p=0.33$
	Video exercise group	45.66 (11.78)	46.66 (13.05)	1.00 (−3.89 to 5.89) $p=0.65$
	Mean difference between groups (95% CI of the difference) *p* value	6.73 (−5.65 to 19.12) $p=0.27$	−5.93 (−9.28 to 21.15) $p=0.42$	
Caregiver Burden Score (out of 88)	Telerehabilitation group	29.70 (15.05)	28.20 (15.28)	1.50 (−0.71 to 3.71) $p=0.16$
	Video exercise group	27.12 (19.69)	26.12 (20.74)	1.00 (−1.36 to −3.36) $p=0.35$
	Mean difference between groups (95% CI of the difference) *p* value	2.57 (−14.75 to −19.90) $p=0.76$	2.07 (−15.90 to 20.05) $p=0.81$	

Table 3 Within group and between group differences between upper extremity muscle strengths in kilograms

	Group	Baseline Mean (SD)	End of treatment Mean (SD)	Mean difference within group (95% CI of the difference) *p* value
Neck flexion	Telerehabilitation	1.69 (0.87)	2.06 (1.09)	− 0.37 (− 0.82 to 0.88) $p=0.10$
	Video exercise	1.03 (0.5)	1.47 (0.57)	− 0.44 (− 0.86 to 0.02) $p=0.40$
	Mean difference between groups (95% CI of the difference) *p* value	0.65 (− 0.04 to 1.36) $p=0.07$	0.58 (− 0.27 to 1.44) $p=0.17$	
Neck extension	Telerehabilitation	2.28 (0.90)	2.93 (0.84)	− 0.65 (− 1.24 to 0.5) $p=0.03$*
	Video exercise	1.87 (0.66)	2.24 (0.61)	− 0.36 (− 1.15 to 0.42) $p=0.31$
	Mean difference between groups (95% CI of the difference) *p* value	0.40 (− 0.37 to 1.17) $p=0.28$	0.68 (− 0.0 to 1.40) $p=0.05$*	
R shoulder abduction	Telerehabilitation	2.82 (0.74)	3.82 (1.29)	− 1.00 (− 1.83 to − 0.16) $p=0.02$*
	Video exercise	1.98 (0.53)	2.28 (0.77)	− 0.30 (− 0.9 to 0.30) $p=0.28$
	Mean difference between groups (95% CI of the difference) *p* value	0.83 (− 0.19 to − 1.46) $p=0.01$*	1.53 (0.48 to 2.57) $p=0.01$*	
L shoulder abduction	Telerehabilitation	2.52 (0.80)	3.53 (1.09)	− 1.01 (− 1.56 to − 0.45) $p=0.03$*
	Video exercise	1.76 (0.62)	2.17 (0.78)	− 0.41 (− 0.89 to 0.06) $p=0.08$
	Mean difference between groups (95% CI of the difference) *p* value	0.75 (0.04–1.45) $p=0.04$*	1.35 (0.42 to 2.28) $p=0.01$*	
R elbow flexion	Telerehabilitation	2.75 (1.38)	3.15 (1.39)	− 0.40 (− 0.84 to 0.46) $p=0.07$
	Video exercise	1.76 (0.52)	2.78 (1.84)	− 0.44 (− 1.21 to − 0.32) $p=0.21$
	Mean difference between groups (95% CI of the difference) *p* value	0.98 (− 0.05 to 2.02) $p=0.07$	0.93 (− 0.26 to 2.13) $p=0.11$	
L elbow flexion	Telerehabilitation	2.78 (1.59)	3.45 (1.59)	− 0.67 (− 1.21 to − 0.12) $p=0.02$*
	Video exercise	1.84 (0.78)	2.28 (1.16)	− 0.44 (− 1.31 to 0.42) $p=0.27$
	Mean difference between groups (95% CI of the difference) *p* value	0.93 (− 0.03 to 2.17) $p=0.13$	1.16 (− 0.26 to 2.13) $p=0.01$*	
R elbow extension	Telerehabilitation	2.59 (1.15)	3.02 (0.99)	− 0.43 (− 1.33 to 0.47) $p=0.33$
	Video exercise	1.62 (0.53)	2.21 (0.87)	− 0.58 (− 1.08 to − 0.9) $p=0.26$
	Mean difference between groups (95% CI of the difference) *p* value	0.96 (0.07 to 1.85) $p=0.03$*	0.80 (− 0.10 to − 1.71) $p=0.01$*	
L elbow extension	Telerehabilitation	2.55 (1.04)	2.85 (1.28)	− 0.30 (− 1.15 to 0.55) $p=0.44$
	Video exercise	1.77 (0.66)	2.02 (0.78)	− 0.24 (− 0.76 to 0.27) $p=0.30$
	Mean difference between groups (95% CI of the difference) p value	0.77 (− 0.08 to − 1.63) $p=0.08$	0.82 (− 0.21 to − 1.87) $p=0.11$	

Table 3 (continued)

	Group	Baseline Mean (SD)	End of treatment Mean (SD)	Mean difference within group (95% CI of the difference) *p* value
R wrist flexion	Telerehabilitation	1.75 (0.69)	2.36 1.15	− 0.61 (− 1.27 to − 0.5) *p* = 0.07
	Video exercise	1.17 (0.36)	1.84 0.64	− 0.66 (− 1.20 to − 0.12) *p* = 0.20
	Mean difference between groups (95% CI of the difference) *p* value	0.57 (0.02 to 1.12) *p* = 0.41	0.51 (− 0.40 to 1.43) *p* = 0.25	
L wrist flexion	Telerehabilitation	1.75 (0.84)	2.15 1.05	− 0.40 (− 0.88 to 0.08) *p* = 0.10
	Video exercise	1.28 (0.42)	1.81 0.47	− 0.52 (− 0.90 to − 0.13) *p* = 0.15
	Mean difference between groups (95% CI of the difference) *p* value	0.46 (− 0.19 to 1.11) *p* = 0.15	0.33 (− 0.46 to 1.14) *p* = 0.38	
R wrist extension	Telerehabilitation	1.82 (0.49)	2.18 1.20	− 0.36 (− 1.18 to 0.46) *p* = 0.34
	Video exercise	1.25 (0.43)	1.95 0.38	− 0.70 (− 1.12 to 0.27) *p* = 0.15
	Mean difference between groups (95% CI of the difference) *p* value	0.56 (0.11 to − 1.01) *p* = 0.17	0.22 (− 0.66 to 1.11) *p* = 0.60	
L wrist extension	Telerehabilitation	1.84 (0.66)	1.96 1.16	− 0.12 (− 0.85 to 0.61) *p* = 0.72
	Video exercise	1.11 (0.33)	1.88 0.33	− 0.77 (− 0.11 to 0.43) *p* = 0.10
	Mean difference between groups (95% CI of the difference) *p* value	0.72 (0.02 to − 1.24) *p* = 0.09	0.07 (− 0.77 to 0.91) *p* = 0.86	

R right, *L* left, *SD* standard deviation

*Marks $p < 0.05$

Previous studies that report an improvement in the function in patients with DMD used cycle-ergometry, which is completely different from the exercise program we implemented in our study. It is a more aerobic-based approach that is harder to implement with video or telerehabilitation [19]. Considering its applicability in a home setting, we chose to use a strengthening-based program. Studies that implemented a similar strengthening program are early studies that have shown that these programs are useful if implemented in the early terms of neuromuscular disorders [20]. Another early study that implemented submaximal exercise in patients with DMD demonstrated that while it has no negative effects, submaximal exercise may not be enough for significant improvements in function [21]. Despite the relatively positive results of these early studies, the data about strength training is quite limited in patients with DMD. A recent study documented that improving muscle strength in patients with DMD is safe in terms of muscle damage and has positive effects on function [22]. However, that study only used the time to ascend and descend stairs for the functional outcome and did not implement other, more global functional measurements. The studies that investigated functional outcomes in a wider manner implemented aerobic exercise training, making the results incomparable to our program [23]. We preferred to implement a more thorough assessment, but improvement in NSAA and QMFT seem to require more than just improvements in muscle strength. A longer follow-up period could show a change in these functional outcome measurements but implementing longer-term therapies can be another challenge in the setting of telerehabilitation. With the current results, it is not possible to deduce any meaningful outcomes without speculation, but a more function-based program can be investigated in future research and can be compared with the more traditional approach that was used in this study. Without the change in function, no change in caregiver burden is an expected outcome, but our results can indicate that the

Table 4 Within group and between group differences between lower extremity muscle strengths in kilograms

	Group	Baseline Mean (SD)	End of treatment Mean (SD)	Mean difference within group (95% CI of the difference) *p* value
R hip flexion	Telerehabilitation	3.25 (1.24)	3.38 (1.40)	− 0.14 (− 1.30 to − 1.02) $p=0.79$
	Video exercise	1.95 (0.80)	2.73 (1.31)	− 0.77 (− 1.56 to 0.01) $p=0.06$
	Mean difference between groups (95% CI of the difference) *p* value	1.29 (0.26 to − 2.32) $p=0.02$*	0.65 (− 0.66 to 1.97) $p=0.30$	
L hip flexion	Telerehabilitation	3.10 (1.14)	3.52 (1.32)	− 0.42 (− 1.32 to − 0.48) $p=0.32$
	Video exercise	2.27 (0.98)	2.51 (1.20)	− 0.23 (− 1.02 to 0.56) $p=0.51$
	Mean difference between groups (95% CI of the difference) *p* value	0.82 (− 0.21 to − 1.85) $p=0.11$	1.01 (− 0.22 to 2.24) $p=0.10$	
R hip abduction	Telerehabilitation	3.62 (1.32)	3.79 (1.08)	− 0.05 (− 0.84 to − 0.74) $p=0.89$
	Video exercise	2.40 (0.91)	2.64 (1.13)	− 0.24 (− 0.78 to 0.29) $p=0.33$
	Mean difference between groups (95% CI of the difference) *p* value	1.22 (0.10 to 2.33) $p=0.04$*	1.14 (0.06 to 2.22) $p=0.03$*	
L hip abduction	Telerehabilitation	3.10 (1.14)	3.56 (1.05)	− 0.05 (− 0.84 to 0.74) $p=0.89$
	Video exercise	2.27 (0.98)	2.90 (1.22)	− 0.36 (− 1.21 to 4.47) $p=0.35$
	Mean difference between groups (95% CI of the difference) *p* value	0.97 (− 0.15 to − 2.11) $p=0.09$	0.66 (− 0.44 to 1.76) $p=0.22$	
R knee flexion	Telerehabilitation	3.14 (1.08)	3.93 (1.17)	− 0.79 (− 1.49 to 0.08) $p=0.03$*
	Video exercise	2.27 (0.78)	3.01 (1.01)	− 0.73 (− 1.73 to 0.27) $p=0.13$
	Mean difference between groups (95% CI of the difference) *p* value	0.86 (− 0.06 to 1.79) $p=0.07$	0.91 (− 0.14 to 1.98) $p=0.05$*	
L knee flexion	Telerehabilitation	2.94 (1.09)	4.03 (1.25)	− 1.09 (− 1.72 to − 0.45) $p=0.01$*
	Video exercise	2.28 (0.92)	3.05 (0.94)	− 0.76 (− 1.58 to 0.05) $p=0.06$
	Mean difference between groups (95% CI of the difference) *p* value	0.46 (− 0.33 to 1.63) $p=0.18$	0.51 (− 0.11 to − 2.05) $p=0.05$*	

Table 4 (continued)

	Group	Baseline Mean (SD)	End of treatment Mean (SD)	Mean difference within group (95% CI of the difference) *p* value
R knee extension	Telerehabilitation	2.98 (1.11)	4.66 (1.50)	− 0.95 (− 1.58 to 0.31) $p = 0.01$*
	Video exercise	2.65 (1.15)	3.46 (1.61)	− 0.81 (− 1.98 to − 0.35) $p = 0.15$
	Mean difference between groups (95% CI of the difference) *p* value	0.32 (− 0.77 to − 1.41) $p = 0.54$	1.19 (− 0.31 to 2.70) $p = 0.05$*	
L knee extension	Telerehabilitation	3.09 (1.00)	4.50 (1.84)	− 1.41 (− 2.75 to − 0.65) $p = 0.04$*
	Video exercise	2.48 (1.01)	3.57 (1.66)	− 1.08 (− 2.38 to 0.21) $p = 0.09$
	Mean difference between groups (95% CI of the difference) *p* value	0.60 (− 0.42 to 1.62) $p = 0.23$	0.92 (− 0.78 to 2.62) $p = 0.05$*	
R ankle dorsiflexion	Telerehabilitation	2.71 (0.86)	3.52 (0.76)	− 0.81 (− 1.45 to 0.16) $p = 0.02$*
	Video exercise	1.97 (0.82)	2.32 (0.64)	− 0.34 (− 0.84 to 0.15) $p = 0.15$
	Mean difference between groups (95% CI of the difference) *p* value	0.73 (− 0.08 to − 1.55) $p = 0.07$	1.19 (0.51 to 1.88) $p = 0.01$*	
L ankle dorsiflexion	Telerehabilitation	2.58 (0.52)	3.12 (0.81)	− 0.54 (− 1.15 to 0.07) $p = 0.01$*
	Video exercise	2.20 (0.95)	2.14 (0.82)	0.05 (− 0.70 to − 0.81) $p = 0.87$
	Mean difference between groups (95% CI of the difference) *p* value	0.38 (− 0.48 to − 1.24) ($p = 0.36$	0.97 (0.17 to − 1.72) $p = 0.02$*	
R ankle plantarflexion	Telerehabilitation	3.12 (1.14)	3.59 (0.94)	− 0.47 (− 1.50 to 0.56) $p = 0.33$
	Video exercise	2.58 (0.80)	2.57 (1.04)	0.01 (− 0.76 to − 0.39) $p = 0.98$
	Mean difference between groups (95% CI of the difference) *p* value	0.53 (− 0.43 to − 1.49) $p = 0.26$	1.01 (0.05 to 1.97) $p = 0.04$*	
L ankle plantarflexion	Telerehabilitation	2.99 (1.63)	3.56 (0.67)	− 0.57 (− 1.81 to 0.67) $p = 0.32$
	Video exercise	2.20 (0.76)	2.50 (1.07)	− 0.30 (− 0.93 to 0.33) $p = 0.30$
	Mean difference between groups (95% CI of the difference) *p* value	0.79 (− 0.47 to 2.05) $p = 0.20$	0.41 (0.20 to − 1.91) $p = 0.02$*	

R right, *L* left, *SD* standard deviation

*Marks $p < 0.05$

telerehabilitation approach does not increase the burden of the caregivers considering their involvement in rehabilitation sessions.

The telerehabilitation approach helps with higher patient compliance when compared to other forms of home-based video exercises due to its ability to involve the caregivers in the process. That is why, in the current COVID-19 pandemic, it gained traction and is being applied to all aspects of health provision. In our study, participation in telerehabilitation sessions was higher than in the video exercise group. We think that the reason for the higher participation rate in the telerehabilitation group is that the telerehabilitation appointments are planned according to the availability of caregivers and physiotherapists, while initially giving videos to the home-based exercise group did not reinforce the scheduled actions. Previous studies showed that caregiver involvement is required in patients with DMD for them to perform their exercises properly [24]. In our study, the exercises were performed by live dialogues with caregivers (video conferencing), explaining the movements, observing and directing them, and giving instructions to correct the movements. However, some tactile applications such as stretching were carried out with the support of caregivers. Some younger patients had problems in focusing on the program, but all the sessions could be completed without major issues. Due to the current situation, we were not able to compare the effects of telerehabilitation with a hands-on rehabilitation approach, so it is not possible to comment on their comparative effectiveness. It is also not possible how the involvement of professionals changes the outcomes. However, our results show that in times where a hands-on approach is not possible, telerehabilitation is better in improving muscle strength than just making patients watch videos and exercise, and the directives from a professional can be just as helpful.

As of winter 2021, the COVID-19 pandemic is still ongoing and has detrimental effects on the physical activity levels of children with physical disabilities, and access to traditional rehabilitation services has become harder for most of these children. These changes have increased the need for telerehabilitation and home-based exercises in all children with disabilities, including patients with DMD [25]. In the study conducted by Tanner et al. during the COVID-19 pandemic, it was stated that telerehabilitation technologies could be used in pediatric rehabilitation, and families reported a high level of satisfaction with this model. However, they noticed limiting factors for implementation, such as technical barriers and privacy concerns [26]. We received mostly positive feedback from caregivers in our study. The caregivers stated that the elimination of the need for transport and the risk of infection were important advantages of telerehabilitation and video exercise programs. Except for some technical issues such as occasional loss of connection, we did not encounter any other problems reported by caregivers during the telerehabilitation sessions.

There are serious limitations of this study. The single-center nature of our study and the involvement of ambulatory patients limited the number of patients included in the study. A multicenter design that includes patients from all functional levels is necessary to make the results more generalizable for all DMD population. Also, the fact that the telerehabilitation application requires an internet connection. The lack of internet connection for some parents has limited the number of our patients, and these approaches can cause the socioeconomically disadvantaged group from receiving necessary therapies. Participation of the video exercise group was based on patient reports and, therefore, could be subjective. Also, video exercise group was unsupervised during their exercise sessions. We did not have an objective measurement of participation such as heart rate monitorization. Last but not least, we could not implement a true control group due to ethical considerations.

Conclusion

A telerehabilitation approach is superior in improving muscle strength to a video-based home exercise, but none of the programs improved functional outcomes in ambulatory patients with DMD.

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s13760-022-01975-4.

Declarations

Conflict of interest The authors declare that they have no conflicts of interest.

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