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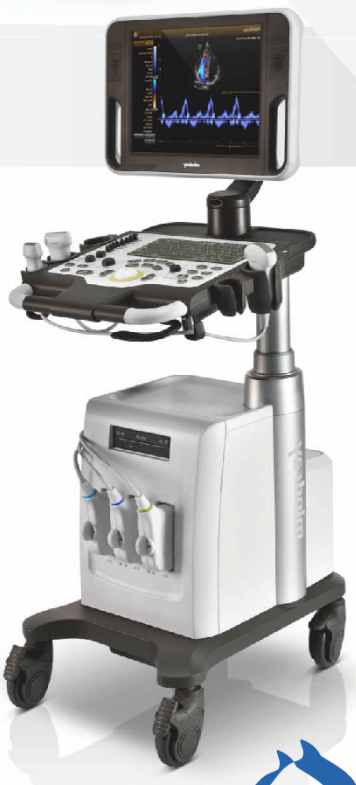

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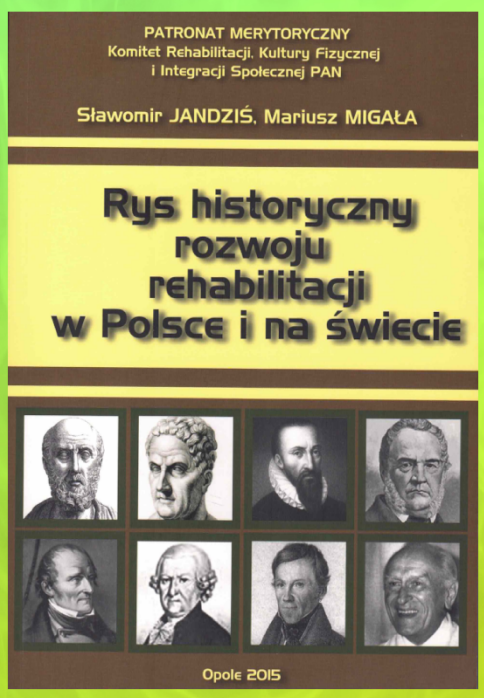

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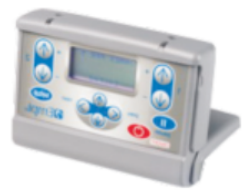

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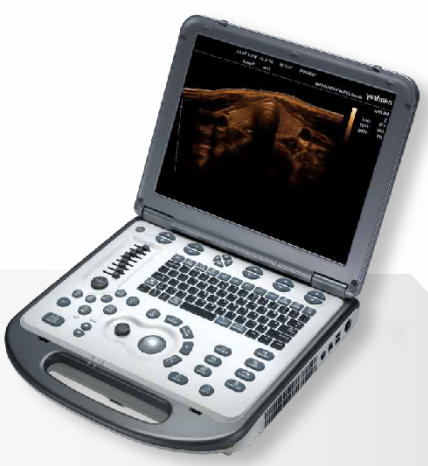

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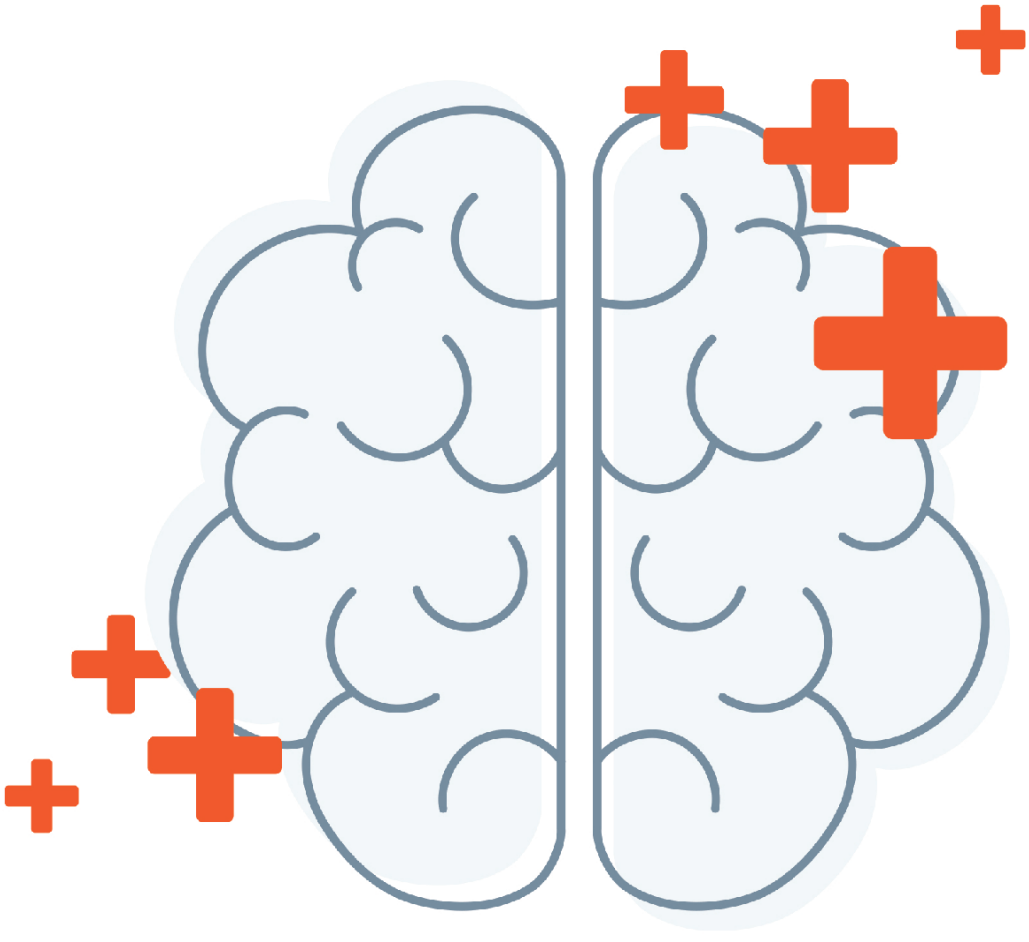

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Effect of respiratory muscle training using Ultrabreathe device on pulmonary function in Duchenne Muscular Dystrophy through telephysiotherapy

Wpływ treningu mięśni oddechowych wykonywanego przy użyciu urządzenia Ultrabreathe na czynność płuc w dystrofii mięśniowej Duchenne'a poprzez telefizjoterapię

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Abstract

Background. Duchenne Muscular Dystrophy (DMD), an X linked disorder resulting in respiratory insufficiency, caused by progressive respiratory muscle weakness particularly in diaphragm. During this pandemic, Telerehabilitation played a major role among patients, especially disabled children. OBJECTIVE: To find out the effect on the pulmonary function in DMD children by training the respiratory muscles by telephysiotherapy. Methodology. Non Experimental design, Case series- pre and post test type. Convenient sampling, sample size was 5. Boys and girls between 9 and 16 years of age were included in the study. Procedure. 5 subjects were selected based on the inclusion and exclusion criteria and were trained for one minute with minimum resistance using ultrabreathe through Telephysiotherapy which is repeated for 3 times in a session with a rest period of 20–30 seconds for six weeks. Outcome measure. Pulmonary Function test. Results. The results of this study shows increased mean value of post test Tidal volume, Forced Vital Capacity, Inspiratory time/Respiration time and Tidal Volume / Inspiration time compared with pre test, whereas other parameters like Duration of Inspiration, Total Lung Volume, Inspiratory Capacity, Forced Expiratory Volume1 does not increased. Conclusion This study concluded that the Duchenne Muscular Dystrophy children improved in Tidal Volume, Forced Vital Capacity, Tidal Volume/Inspiration Time, and Inspiration Time/Respiration Time whereas all other parameters such as Duration of Inspiration, Total lung volume, Inspiratory capacity and Forced Expiratory volume1 maintained after 6 weeks of respiratory muscles training with ultrabreathe device.

Keywords

Duchenne Muscular Dystrophy, Telephysiotherapy, Pulmonary Function Test, Ultrabreathe Device

Streszczenie

Wprowadzenie. Dystrofia mięśniowa Duchenne'a (DMD), zaburzenie sprzężone z chromosomem X, powodujące niewydolność oddechową, spowodowaną postępującym osłabieniem mięśni oddechowych, zwłaszcza przepony. W czasie pandemii telerehabilitacja odegrała istotną rolę wśród pacjentów, zwłaszcza niepełnosprawnych dzieci. Cel: Zbadanie wpływu treningu mięśni oddechowych za pomocą telefizjoterapii na czynności płuc u dzieci z DMD. Metodologia. Projekt nieeksperymentalny, badanie przypadku - przed i po zastosowaniu treningów. Próba okolicznościowa, wielkość próby wynosiła 5. Do badania włączono chłopców i dziewczęta w wieku od 9 do 16 lat. Procedura. W badaniu wzięło udział 5 osób wybranych na podstawie kryteriów włączenia i wyłączenia. Uczestnicy wykonywali trening przez jedną minutę przy minimalnym oporze przy użyciu urządzenia Ultrabreathe, otrzymując instrukcje w ramach telefizjoterapii, realizowanej w następujący sposób: trzy serie na sesję, z czasem na odpoczynek 20–30 sekund, przez sześć tygodni. Miary wyników. Badanie funkcji płuc. Wyniki. Wyniki tego badania po zastosowanym treningu pokazują zwiększoną średnią wartość objętości oddechowej, natężonej pojemności życiowej, czasu wdechu/czasu oddychania i objętości oddechowej/czasu wdechu w porównaniu z wynikami wstępnymi, podczas gdy inne parametry, takie jak czas trwania wdechu, całkowita objętość płuc, pojemność wdechowa, natężona objętość wydechowa pierwszosekundowa nie zwiększyły się. Wniosek. W badaniu tym stwierdzono, że u dzieci z dystrofią mięśniową Duchenne'a objętość oddechowa, natężona pojemność życiowa, objętość oddechowa/czas wdechu oraz czas wdechu/czas oddychania poprawiły się, podczas gdy wszystkie inne parametry, takie jak czas trwania wdechu, całkowita objętość płuc, pojemność wdechowa i natężona objętość wydechowa pierwszosekundowa utrzymują się na tym samym poziomie po 6 tygodniach treningu mięśni oddechowych na urządzeniu Ultrabreathe.

Słowa kluczowe

dystrofia mięśniowa Duchenne'a, telefizjoterapia, test czynnościowy płuc, urządzenie Ultrabreathe

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Introduction

Duchenne Muscular Dystrophy (DMD), an X linked disorder resulting in respiratory insufficiency, caused by progressive respiratory muscle weakness particularly in diaphragm. The chromosomal abnormality leads to the inadequate function of the dystrophin protein and it is one of the most prevalent and severe muscular dystrophies [1, 2, 3]. The mutations in the DMD gene, there is dystrophin protein insufficiency which producing insufficient muscle responses to mechanical stretching and contraction, gradual muscle fiber degradation, resulting in progressive muscular dysfunction [4, 5].

Duchenne Muscular Dystrophy (DMD) had a high prevalence of 0.9 to 16.8 occurrences per 100,000 males, from newborns to the oldest living people. The overall prevalence of DMD in the normal community (males and females) ranged from 0.7 to 7.7 incidences for each 100,000, with Sweden having the least value and Egypt having the largest value. 13 (59.1%) were Europeans, 4 (18.2%) were Americans, 2 (9.1%) were Asians, and 3 (13.6%) were Africans. The global birth prevalence as a whole was 20.5 cases per 100,000 males. This disorder affects one out of every 5000 boys causing proximal muscular weakness in early childhood [6]. The pulmonary deficiency and respiratory infection in DMD, causes about 10% of deaths. Children with Duchenne Muscular Dystrophy use their upper limb to "walk" and initiate movement from a squatting position, due to proximal muscle weakness and a lack of hip and thigh muscle strength. Gross motor delay, waddling gait, difficulty rising from the ground, calf pseudo-hypertrophy, and increased curvature of lumbar spine and frequent falls are common symptoms of DMD. Elevated serum creatine kinase (CK) levels or an inadvertent detection of increased hepatic transaminases are less common manifestations [3, 7, 8]. Clinically cardiomyopathy appears after the age of ten, affects one-third of individuals by the age of fourteen, and affects all children after the age of eighteen [9]. Despite the high prevalence of cardiac involvement, most boys are asymptomatic as a result of their lack of physical activity. Fatigue and respiratory failure are caused by the difference between the mechanical load and the capacity of the respiratory muscle [10, 11]. Respiratory deficit in Duchenne Muscular Dystrophy is caused by progressive respiratory muscle weakness, particularly in diaphragm [12]. The ability to cough, speak, and swallow is also impacted by a reduction in respiratory muscle strength. The capacity of the lungs is improved by increasing respiratory muscle strength, which also helps to manage an increase in respiratory load. Bronchial hygiene, which is crucial to reducing the incidence of respiratory infections, is maintained by respiratory muscle strength in addition to maintaining gaseous exchange and pulmonary functions [13, 14].

Many rehabilitation programme have been halted as a result of the pandemic (COVID-19 epidemic). Duchenne Muscular Dystrophy children encounter the problem of functional decline in this unique condition. In addition, the social isolation accompanied by quarantine could trigger a variety of psychological issues, including acute stress disorders, irritability, sleeplessness, distress, and mood disturbances. Long-term isolation is associated with a high degree of stress that leads to psychological distress, and severe anxieties [6]. During this pandemic Telerehabilitation plays a major role in patients, especially in disabled children. Telerehabilitation research aims to reduce the cost of telerehabilitation services while also optimizing the usage of telerehabilitation technology in a variety of situations. These objectives are attainable, especially given the rising body of research that supports the practicality, acceptability, and efficacy of numerous digital treatments [15].

The technology employed by rehabilitation specialists can range from simple day-to-day applications (e.g., contact via phone calls or email) to complex technologies (e.g., specialized equipment deployed in a clinical environment and at home). When the focus of the therapies is on helping the children and their families, problem-solving with them to nurture the child's development and functioning, telerehabilitation may be particularly well adapted to implementing best practices for children with disabilities [16].

Telephysiotherapy has proved effective in the home management of neuromuscular disease (NMD) patient [12, 16]. Parents has been oriented about the usage of telephysiotherapy before the treatment protocol.In this study, the respiratory muscles were trained by an effective respiratory muscle trainer named Ultrabreathe. It works on the principle of resistance. It gives easy and convenient means of increasing the strength and the endurance of the respiratory muscles.

The goal of the study is to show an effect of Ultrabreahe device on the Pulmonary Function in Duchenne Muscular Dystrophy children by training the respiratory muscles by telephysiotherapy.

Materials and methodology

The study was approved by the Institutional Ethical Committee of SRM Medical College and Hospital and Research Center. Ethical Clearance Number: 2889. An Experimental study was done with convenient sampling on 5-wheel chair depended boys and girls diagnosed with DMD of age between 9–16 years and their total lung capacity ranges from 0.9 l to 4.2 l were included. And those boys and girls with cardiac abnormalities, lower respiratory tract infections were excluded. Followed by the spirometer analysis of lung volume their respiratory muscles were trained using Ultrabreathe device with minimal resistance set by adjusting the cap. Telephysiotherapy was conducted through the parent’s mobile via whattsapp application. It was conducted thrice a week, and the procedure was repeated for 3 times a session and 2 sessions per day i.e morning and evening. First demonstration was given to the parents during the pre test of spirometric measurement. Parents were oriented about the usage of telephysiotherapy before the treatment was started. They were instructed to inhale to the maximum for three times a session with rest time of 20–30 seconds in between. The patient is expected to maintain a upright sitting posture throughout the procedure. The mouthpiece of Ultrabreathe were sterilized at the end of each session. At the end of six weeks spirometer analysis were conducted to assess their lung volume. The patient was instructed to stop the exercise if any discomfort is experienced. Spirometry tests are used to evaluate pulmonary function, such as lung capacity. Slow Vital Capacity (SVC) is a breathing technique in which the child inhales completely before gently exhaling all of the air in the lungs. This is referred to as IVC (Inspiratory Vital Capacity). We can determine Inspiratory Reserve Volume, Tidal Volume, Duration of Inspiration,

Tidal Volume Per Inspiration Time, Inspiratory Time, and Inspiratory Capacity using Slow Vital Capacity.Forced Vital Capacity (FVC) is a spirometer test in which a child is required to exhale as hard and fast as possible. Forced Expiratory Volume in One Second refers to the volume that expired in the first second of the FVC test (FEV1). FEV1 percent is calculated by multiplying FEV1 by the forced vital capacity (FVC)*100.

Results

The collected data was tabulated and the data was analyzed.

Table 1 shows the demographic data for DMD children.
Table 2 shows the pre test value of Pulmonary Function Test i.e., Tidal Volume, Total Lung Volume, Inspiratory Capacity, Forced Vital Capacity, Forced Expiratory Volume 1 sec, Inspiratory Time/Respiration Time, Tidal Volume/Inspiration time, Duration of Inspiration in DMD Children.
Table 3 shows post test value of Pulmonary Function Test in DMD Children. The mean value has increased in Tidal Volume, Forced Vital Capacity, Inspiratory Time/Respiration Time, and Tidal Volume/Respiration Time from pre test to post test.

Table 1. Demographic data for DMD children

Case No.	Age	Gender
Case 1	9	Girl
Case 2	12	Boy
Case 3	13	Girl
Case 4	15	Boy
Case 5	16	Boy

Table 2. Pre test value of pulmonary function test in DMD Children

Case No.	Age	TV (LI)	TLV (L)	IC (L)	FVC (L)	FEV1 (L)	IT/RT	TV/IT	D OF I (sec)
Case 1	9	1.41	2.62	2.02	1.77	1.7	0.4	0.75	1.92
Case 2	12	1.52	2.22	1.84	1.5	1.26	0.44	0.7	2.17
Case 3	13	1.4	2.64	2.02	1.77	1.68	0.39	0.75	1.91
Case 4	15	0.69	0.98	0.84	0.82	1.71	0.49	0.34	2.1
Case 5	16	1.18	2.02	1.72	0.47	1.34	0.48	0.62	2.02
Mean	13.0	1.24	2.096	1.68	1.26	1.53	0.44	0.632	2.02

Table 3. Post test value of pulmonary function test in DMD Children

Case No.	Age	TV (LI)	TLV (L)	IC (L)	FVC (L)	FEV1 (L)	IT/RT	TV/IT	D OF I (sec)
Case 1	9	1.54	2.04	1.8	1.74	1.53	0.5	1.15	1.35
Case 2	12	1.53	1.74	1.66	1.61	1.44	0.43	0.7	2.19
Case 3	13	1.49	2.05	1.78	1.74	1.52	0.49	1.15	0.77
Case 4	15	1.04	1.14	1.12	0.6	1.52	0.76	0.7	1.51
Case 5	16	1.29	2.34	2.04	1.59	1.13	0.56	0.87	1.7
Mean	13.00	1.37	1.86	1.68	1.45	1.42	0.54	0.91	1.50

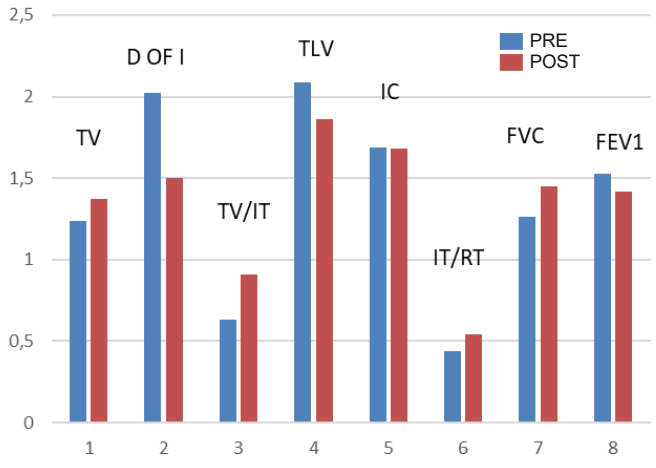

Figure 1. Comparing the pre and post test values of pulmonary function test in DMD Children

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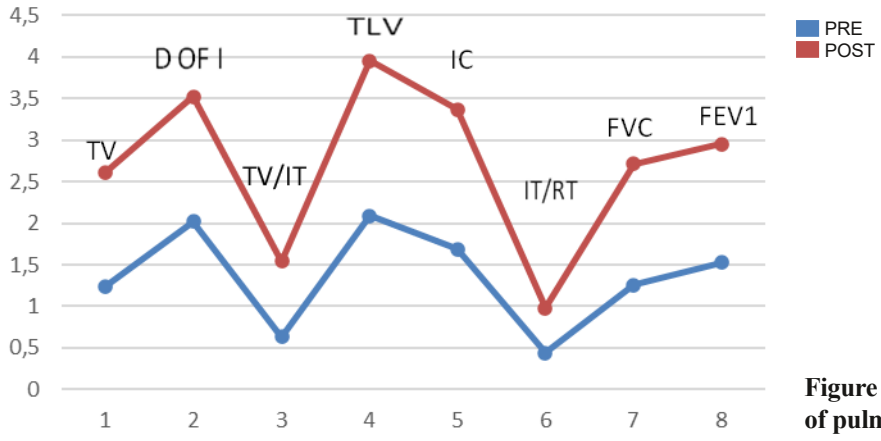

Figure 2. Comparing the pre and post test values of pulmonary function test in DMD Children

Discussion

The purpose of this study is to train the respiratory muscle using Ultrabreathe device through Telephysiotherapy in Duchenne Muscular Dystrophy children.To our knowledge, this is the first study to examine the pulmonary function of DMD children by respiratory muscle training by Ultrabreathe device through Telephysiotherapy. The mean value of post test of Tidal Volume is increased when compared with pretest value of Tidal Volume where its normal value is 0.5L. The mean value of posttest of Tidal Volume/Inspiration time is increased when compared with pretest value of tidal /Inspiration time where its normal value is 1.25. The mean value of post test of Forced Vital Capacity is increased when compared with pre test value of Forced vital capacity where its normal value is 1.22. The mean value of posttest of Inspiratory time / Respiration time is increased when compared with pretest value of Inspiratory time / Respiration time where its normal value are 0.1.

Malarvizhi D et.al., (2022) Tidal Volume only was improved as a result of inspiratory muscle training utilizing the device on pulmonary function in DMD children, with no significant improvement in the other components [17], Which correlates with this study were respiratory muscle training was done using ultrabreathe device through telephysiotherapy, the average value of tidal volume, forced vital capacity, tidal volume/ inspiration time and inspiratory time / respiration time shows improvement whereas other parameters like duration of inspiration, total lung volume, inspiratory capacity does not show much improvement. According to Hallett S et.al., (2020) physiological demands increased minute ventilation, resulting in an increase in oxygenation levels, which led to an increase in both tidal volume and inspiratory rate during exercise. After 6 weeks of training, the tidal volume has increased considerably and because of the rise in Tidal volume, a significant increase in minute ventilation is expected resulting in increased oxygenation [18].

As a result of the pandemic, Duchenne Muscular Dystrophy interdisciplinary healthcare system had to be reorganise, Telephysiotherapy acts as an important tool for physiotherapists to train children with DMD without them needing to visit the physiotherapy department. For the time period, telemedicine options appear to be the most effective means of offering rehabilitation services.

Before the treatment procedure, parents were educated on how to use Telephysiotherapy and the Ultra breathe device. Parents were really excited and interested to participate in the study after hearing about the need for it and how it would benefit their children because there is a lack of physical activity and physiotherapy treatment sessions owing to the pandemic. Teaching children how to use the device was difficult as some children did not understand how to use it. In these circumstances, caregivers/parents assisted the children on how to use the device.

Parents/caregivers of children with DMD who participated in the study asked if the device can be regularly used as a home exercise program for the child because it motivates the children throughout the study, which keeps them physically active, Agnieszka Sobierajska-Rek (2021) stated that children can continue their home- based therapy with the support of caregivers and physiotherapist supervision (online communication or video). Caregivers of Duchenne muscular dystrophy children prefer online videos/instructions/video guidance to live workshops [1].

Most of the parents/caregivers were satisfied with telephysiotherapy session than in person service where their child are actively participated and took some time to focus in telephysiotherapy training programme, Aashaka C Shah et.al (2021) also concluded that the telemedicine services are comparable to or better than in-person services. Children and caregivers have also consistently reported being satisfied. Telemedicine visits are more satisfying than in-person encounters. This indicates potential for telemedicine in paediatric settings, particularly when times when social distance is required, such as when the outbreak of COVID-19 [15]. It is seen that the Duration of Inspiration, Total Lung Volume, Inspiratory Capacity and Forced Expiratory Volume 1 maintained after 6 weeks of treatment, Which is in accordance with the study done by Frederic Proulx et.al, (2021) where heconcluded that regular Lung Volume Recruitment /airway clearance/Pulmonary Function Test for a longer duration will improve the lung volume capacity and maintain respiratory status [19].

Orlando E. Flores G et. al., (2009) has showed that strengthening the inspiratory muscle for nine weeks can significantly enhance respiratory muscle endurance in people with DMD. The observation of peak coughs flow during long periods of

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muscle training confirms that DMD children respiratory muscle endurance and force can be increased by undergoing long periods of muscle training [5, 14], which correlates with this study where the Respiratory muscle training with Ultrabreathe device for a longer duration can also improve pulmonary function status in Duchenne Muscular Dystrophy children. Rasha Saadi Abbas et.al, (2020) stated that the study did not find a significant difference between blood group phenotype and asthma severity regarding forced expiratory volume in one second/ forced vital capacity ratio (FEV1/FVC) [20].

This research result goes in hand with Aldrich et.al, (1989) revealed that training the respiratory muscles for a long period of time can improve treatment efficacy in DMD and also concluded that inspiratory resistance training combined with concurrent delivery of intermittent mandatory ventilation increased the effectiveness in patient with DMD [11]. Hence it can be inferred that due to short duration of the treatment session and the disease is progressive in nature, there is no marked improvement in this study. The lack of changes in Duration of Inspiration, Total Lung Volume, Inspiratory Capacity, and Forced Expiratory Volume in One Second (FEV1) after Ultrabreathe treatment in DMD children was due to the climatic conditions, as the treatment was administered during the winter season. Another reason for the lack of changes in the components is the fact that they were only treated for six weeks. If they are treated for an extended period of time, the prognosis may be considerable.

Conclusion

This study concluded that the Duchenne Muscular Dystrophy children improved in Tidal Volume, Forced Vital Capacity, Tidal Volume/Inspiration Time, and Inspiration Time/Respiration Time whereas all other parameters such as Duration of Inspiration, Total lung volume, Inspiratory capacity and Forced Expiratory volume1 maintained after 6 weeks of respiratory muscles training with Ultrabreathe device.

The limitations of this study is the study duration was shorter and the sample size was very small was included and recommendations of the study longitudinal studies can be implemented in future research and respiratory devices other than PFT can be correlated in future research.

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