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Effects of Five-month Training of Playing Harmonica on Pulmonary Function in Patients With Neuromuscular Disease: A Pilot Study

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Abstract

Background: Progressive muscle weakness is aggravated not only in the skeletal muscles but also in the respiratory muscles in many patients with neuromuscular diseases (NMD). Inspiratory muscle training (IMT) has been reported as therapy for pulmonary rehabilitation to improve respiratory strength, endurance, exercise capacity, and quality of life, and to reduce dyspnea.

Objects: The purpose of this study was to determine the effect of playing harmonica for 5 months on pulmonary function by assessing the force vital capacity (FVC), peak cough flow (PCF), maximal inspiratory pressure (MIP), maximal expiratory pressure (MEP), and maximal voluntary ventilation (MVV) in patients with NMD.

Methods: Six subjects with NMD participated in this study. The subjects played harmonica once a week for 2 hours at a harmonica academy and twice a week for 1 hour at home. Thus, training was performed thrice a week for 23 weeks. The examiner assessed pulmonary function by measuring FVC in the sitting and supine positions and PCF, MIP, MEP, and MVV in the sitting position at the beginning of training and once a month for 5 months.

Results: Both sitting and supine FVC significantly increased after playing harmonica (p=.042), as did MIP (p=.043) and MEP (p=.042).

Conclusion: Playing harmonica can be used as an effective method to improve pulmonary function in patients with NMD.

Keywords: Harmonica; Neuromuscular disease; Pulmonary function test.

Introduction

Neuromuscular diseases (NMD) are characterized by progressive weakness of respiratory, skeletal, and bulbar muscles, as well as myopathies and cardiac muscle weakness (Griggs et al, 1981). Pulmonary complications lead to increase in mortality and morbidity in patients with NMD (McCool and Tzelepis, 1995). Patients with NMD are noted to have an advance restrictive pulmonary disease pattern due to the progressive weakening of the respiratory muscles. Patients with NMD have severe expiratory and inspiratory muscle insufficiency that decreases tidal volumes, sighing, and coughing those results in reduction in lung insufflation and thoracic chest wall (McCool et al, 1986). That symptom leads to ineffective coughing, decreased respiratory muscle power, and decreased vital capacity during otherwise benign chest infections (Bach et al, 1998; Gibson et al, 1977).

A previous study has reported on the benefits of moderate-resistance training in progressive NMD for respiratory muscle power and especially reported that training for 12 weeks or longer would be needed to increase maximal expiratory pressure (MEP) or max-

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Table 1. Characteristics of the six subjects with neuromuscular disorders

Gender	Age	Scoliosis	Diagnosis	Height(㎝)	weight(㎏)	Functional status
Male	38	X	MD[a]	163	65	Wheelchair
Male	56	X	BMD[b]	168	92	Wheelchair
Female	44	X	Myopathy	164	52	Wheelchair
Male	48	X	Myopathy	180	69	Wheelchair
Male	21	O	DMD[c]	152	47	Wheelchair
Female	28	O	PMD[d]	148	66	Wheelchair

[a]muscular dystrophy, [b]Becker muscular dystrophy, [c]Duchenne muscular dystrophy, [d]progressive muscular dystrophy.

imal inspiratory pressure (MIP) (Aitkens et al, 1993). In many patients with NMD patients, progressive muscle weakness is aggravated not only in the skeletal muscles, but also in the respiratory muscles. Inspiratory muscle training (IMT) has been reported as therapy for pulmonary rehabilitation to improve respiratory strength, endurance (Dimarco et al, 1985), exercise capacity, and quality of life, and to reduce dyspnea (Lötters and Kwakkel et al, 2002; Weiner and McConnell, 2005). In the early stages of Duchenne muscular dystrophy (DMD), IMT with pressure threshold device was more effective than other IMT devices when the forced vital capacity (FVC) is preserved (Kang et al, 1998). Other studies have also proven that expiratory and inspiratory muscle endurance can be improved by specific exercises involving resistant inspiratory breathing in patients with various types of nonmyotonic muscular dystrophies (Dimarco et al, 1985; Kang et al, 1998; McCool and Tzelepis, 1995).

Playing harmonica has an effect similar to IMT based on the similarity of performing inspiration and expiration through a device that provides resistance with nose clip (due to the size of the holes in the harmonica), and provides an advantage similar to IMT (Alexander and Wagner, 2012). In addition, playing harmonica may be enjoyable, user-friendly, and useful for improving their breathing dynamics (Jeffery et al, 2012). A previous study has reported on the benefits of However, in the present, no studies have reported a therapeutic effect of playing harmonica in patients with NMD.

Therefore, the purpose of this study was to determine whether 5-month training of playing harmonica can affect pulmonary function by assessing FVC, peak cough flow (PCF), MIP, MEP and maximal voluntary ventilation (MVV) in patients with NMD. We hypothesized that FVC, PCF, MIP, MEP, and MVV would increase after 5 months of harmonica playing in patients with NMD.

Methods

Subjects

G*power software (G*power software 3.1.2; Franz Faul, University of Kiel, Kiel, Germany) was used for power analysis. From the data of a pilot study of eight subjects, the necessary sample size was three subjects, with a significance level of .05, power of .8, and effect size of 5.5. The subjects from the Korean Muscular Dystrophy Association volunteered to participate in the study. Finally, six subjects with NMD were selected from eight subjects of the pilot study. Two subjects dropped out from the study because of deteriorating general medical condition, such as pneumonia and facial muscle weakness. The characteristics of the six subjects with NMDs are presented in Table 1. NMD diagnosis was established by standard criteria (Emery et al, 1997). We evaluated six subjects who were diagnosed with NMD based on case history, clinical electromyogram, and muscle biopsy. Exclusion criteria include those who refused to undergo the test for maneuvers, those

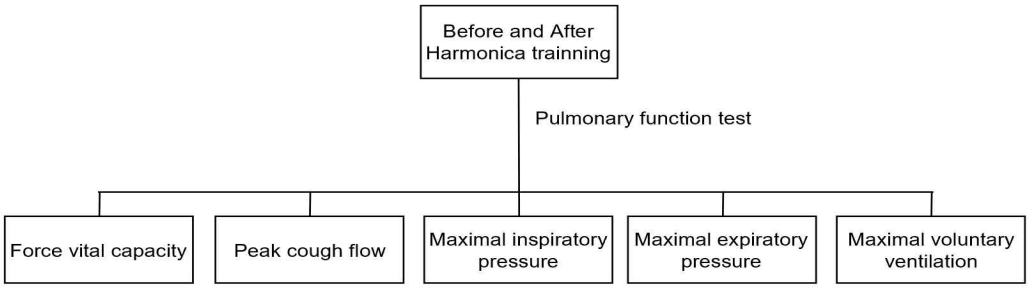

Figure 1. Flow chart of Measurement procedure.

with acute pulmonary disease conditions (e.g. pneumonia, pneumothorax, etc.), and those with an indwelling tracheostomy tube. The subjects with inability to cooperate due to mental retardation were also excluded for this study. Before the measurements, the subjects were informed of the experimental protocol. The subjects read and signed an informed consent form before participation. This study was approved by the Gang-Nam Severance Hospital Institutional Review Board (approval number: 3-2017-0137).

Harmonica playing

All subjects used the same 22-hole harmonica (Weissenburg, 2202F, Taichung, Taiwan). The cover plates of the harmonica were made of vacuum titanium. The subjects used a harmonica holder as an adjunctive tool. All subjects played the harmonica in the harmonica academy of the Korean Muscular Dystrophy Association with and without an instructor at home. The subjects played harmonica once a week for 2 hours at the harmonica academy and twice a week for 1 hour at home. Thus, training was performed thrice a week for 23 weeks.

Measurement procedure

The subjects were assessed for pulmonary function using routine pulmonary function test and continuous routine check of FVC, PCF, MIP, MEP, and MVV at the beginning of training, after the end of the 5-month training. Pulmonary function test was performed in sitting position. The FVC tests were performed in both sitting and supine positions. In the supine position, the abdominal contents exert an upward pressure on the diaphragm, which pushes the thoracic cavity, thereby decreasing the FVC. All pulmonary function data were collected in each subject's hospital room. The subjects performed the tests at least thrice, and resting time was given. The highest record was selected (Won et al, 2015).

Forced vital capacity

FVC was measured in both the sitting and supine positions by using a spirometer (CareFushion, Micro TM Spirometer, Kent, UK). Each subject sat in a wheelchair with back support and lay on the bed for the supine position. The subjects inhaled as deeply as possible, and then maximally exhaled into the spirometer. We calculated the predicted FVC values (FVC pre) based on age, height, and weight (Wilson et al, 1984). The relative FVC values were reported as FVC/FVC pre (%).

Peak cough flow

PCF was measured by a peak flow-meter (Health Scan Products Inc., Assess Peck Flowmeter, NJ, USA). The subjects were instructed to take the deepest breath and then to cough as strongly as possible through a peak flow-meter (Park et al, 2010).

Maximal inspiratory pressure and maximal expiratory pressure

Maximal respiratory pressure and reflecting pulmonary muscle strength were measured using

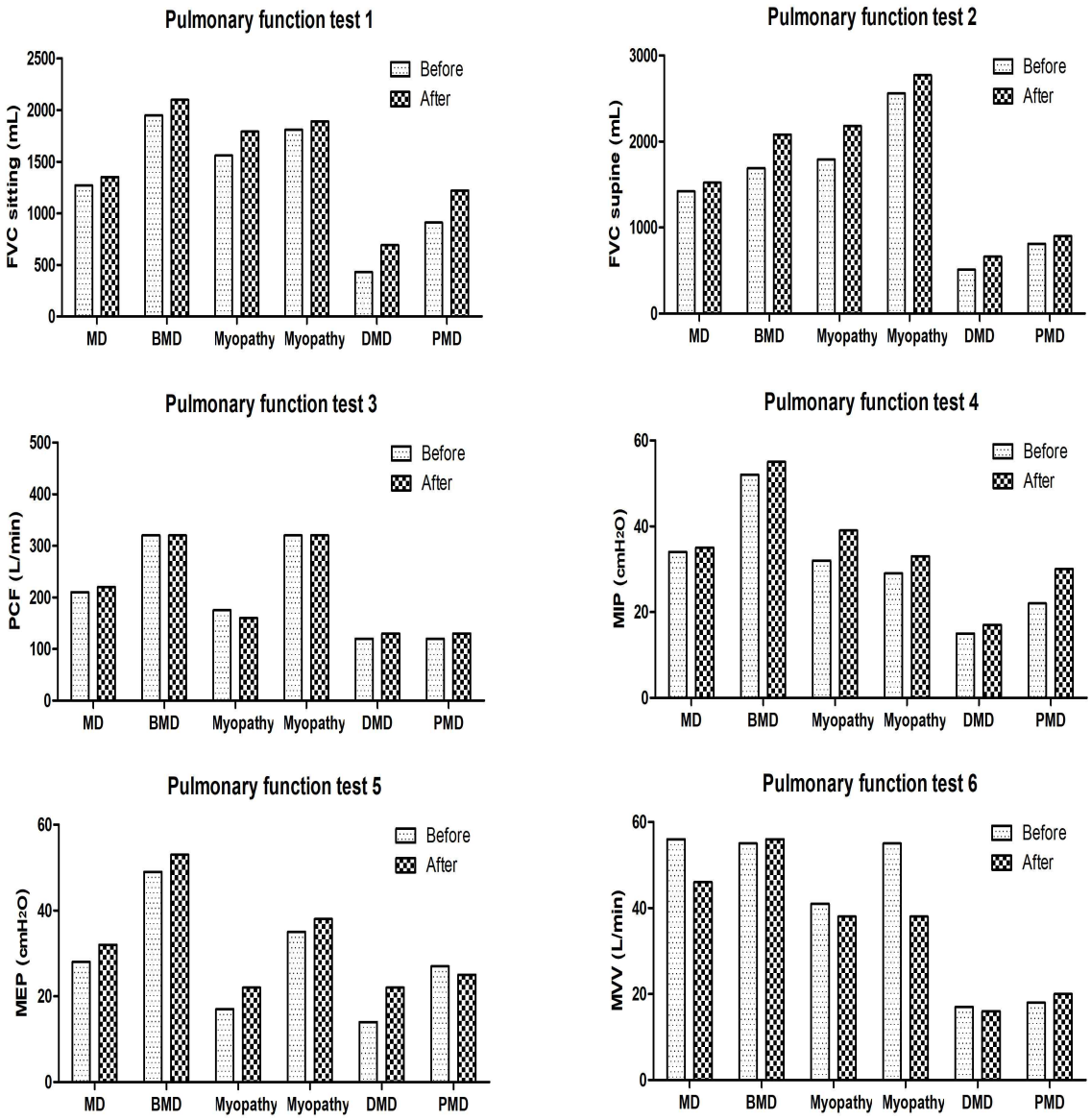

Figure 2. Before and After of pulmonary function measurement (BMD: Becker muscular dystrophy, DMD: Duchenne muscular dystrophy, MD: muscular dystrophy, PMD: progressive muscular dystrophy FVC: forced vital capacity, PCF: peak cough flow, MIP: maximal inspiratory pressure, MEP: maximal expiratory pressure, MVV: maximal voluntary ventilation).

mouth pressure (CareFushion, Micro RPMTM, Hoechberg, Germany) in a sitting position. To measure MEP, the subjects performed maximal expiratory effort after maximal inspiration. MIP was measured by exerting maximal inspiratory effort after maximal expiration. A conventional nose clip and mouthpiece were used to prevent air leakage. If the subjects have difficulty in sealing their mouths, the examiner helped sealing the subject's mouth firmly around the mouthpiece. To measure these pressures, effort was maintained for at least 1 second. We calculated the predicted MEP (MEP pre) and MIP (MIP pre) values based on age, height, and weight (Wilson et al., 1984; Won et al., 2015). The relative MEP and MIP

values were presented as MEP/MEP pre (%) and MIP/MIP pre (%).

Maximal voluntary ventilation

MVV is an index of pulmonary function variable used to determine respiratory muscle endurance (Kor et al, 2004). The traditional intervention of measuring MVV was to let the subject breathe the largest ventilation volume that could be breathed into and out of the lungs during a 12-second interval with maximal voluntary effort. The subjects inhaled deeply (with a volume greater than the tidal volume but lower than the FVC) and rapidly for a 12-second interval, with ventilation flow measured using a pony FX (COSMED, Pony FX, Rome, Italy).

Statistical analysis

Because the sample size was small, the Wilcoxon signed-rank test was used to compare pulmonary functions (FVC, PCF, MIP, MEP and MVV) between the beginning of training and after the end of the 5-month training. Statistical significance was set at .05. All data were analyzed using statistical package for the SPSS ver. 23.0 (IBM corp., Armonk, NY, USA).

Results

Table 2 and Figure 2 shows the collected data of each patient during pulmonary function test. Table 3 shows the pulmonary function measurement results, comparisons of before and after playing harmonica. Both sitting and supine FVC significantly increase after playing harmonica (p= .042), as well as MIP (p= .043) and MEP (p= .042). Both PCF and MVV did not significantly increase after playing harmonica (p=1.000, p=.080, respectively).

Discussion

The purpose of this study was to determine the effect of 5-month training of playing harmonica on pulmonary function by assessing FVC, PCF, MIP, MEP, and MVV in patients with NMD. This was the first study to objectively assess the effect of playing harmonica on pulmonary function in patients with NMD. Our result partially supported the research hypothesis. Playing harmonica was effective in increasing FVC, MIP, and MEP in patients with

Table 2. Evaluation of pulmonary function measurement

Diagnosis	TEST	FVC[a] sitting(㎖)	FVC supine(㎖)	PCF[b] (L/min)	MIP[c] (cmH_2O)	MEP[d] (cmH_2O)	MVV[e] (L/min)
MD[f]	Before	1270	1420	210	34	28	56.3
	After	1350	1520	220	35	32	46
BMD[g]	Before	1950	1690	320	52	49	54.8
	After	2100	2080	320	55	53	55.6
Myopathy	Before	1560	1790	175	32	17	40.5
	After	1790	2180	160	39	22	38
Myopathy	Before	1810	2560	320	29	35	54.8
	After	1890	2770	320	33	38	53
DMD[h]	Before	430	510	120	15	14	16.7
	After	690	660	130	17	22	15.7
PMD[i]	Before	910	810	120	22	27	18.1
	After	1220	900	130	30	25	20.3

[a]forced vital capacity, [b]peak cough flow, [c]maximal inspiratory pressure, [d]maximal expiratory pressure, [e]maximal voluntary ventilation, [f]muscular dystrophy, [g]Becker muscular dystrophy, [h]Duchenne muscular dystrophy, [i]progressive muscular dystrophy.

Table 3. Pulmonary function test result

	Z	Md[a]5 - Md1	p
FVC[b]_sitting5[c] - FVC_sitting1[d]	-2.032	155.00	.042*
FVC_supine5 - FVC_supine1	-2.032	245.00	.042*
PCF[e]5 - PCF1	.000	-2.50	1.000
MIP[f]5 - MIP1	-2.023	3.50	.043*
MEP[g]5 - MEP1	-2.032	1.00	.042*
MVV[h]5 - MVV1	-1.753	-5.65	.080

[a]median, [b]forced vital capacity, [c]before 5-month training, [d]after 5-month training-5, [e]peak cough flow, [f]maximal inspiratory pressure, [g]maximal expiratory pressure, [h]maximal voluntary ventilation, *p<.05.

NMD. The reason this study set the experiment period as 5 months was that no study has been conducted on harmonica training for patients with restrictive lung disease, while previous research findings suggested that training for ≥12 weeks would be needed to improve the pulmonary function of patients with neuromuscular disease (Aitkens et al., 1993). The training period was determined as the longer period of 5 months.

After a 5-month training, FVC was significantly greater in both the sitting and supine positions. Sitting FVC increased by 12%, and supine FVC increased by 18%. These findings support our research hypothesis. Both deep breathing exercises and playing harmonica produced comparable results although both exercises are different (Jeffery et al, 2012). A previous study indicated that deep breathing exercise increased FVC in patients with NMD (Adams and Chandler, 1974). Playing harmonica might be similar to deep breathing exercises in increasing VC (Jeffery et al, 2012). Previous studies have reported that FVC was greater in the sitting position than in the supine position because the diaphragm maintains as a primary inspiratory muscle in patients with NMD (Mcdonald et al, 1995; Park et al, 2010). However, in this study, interestingly FVC was greater in the supine position than in the sitting position in two subjects. This result could be because the two subjects had scoliosis, and the scoliotic curve and rib cage malposition might reduce FVC in the sitting position. Decreased FVC in the sitting position can also occur in patients with NMD (Park et al, 2010). Thus, playing harmonica might be useful to improve both sitting and supine FVC in patients with NMD.

Our results showed that MIP and MEP significantly increased after training. MIP and MEP increased by 10% and 11%, respectively. These findings support our research hypothesis. Our result is consistent with the finding of previous studies that investigated the effects of IMT on MIP and MEP (Ansved, 2001; Jeffery et al., 2012; Kang et al, 2006a). The only difference between previous studies and our study was that we used playing harmonica as an independent variable to increase MIP and MEP. In previous studies, MIP increased after the training when patients with NMD used IMT with pressure threshold device for respiratory muscle training (Kang et al, 2006a; McCool and Tzelepis, 1995). Moreover, playing harmonica was an effective therapy to improve breathing, similar to IMT (Jeffery et al, 2012). Most of the muscle endurance studies have reported increased MIP instead of MEP (Kang et al, 2006a; McCool and Tzelepis, 1995), but playing harmonica improved not only MIP, but also MEP in our study. The possible reason for improvement in MIP and MEP may be because playing harmonica played a crucial role similar to IMT and expiratory muscle training. Respiratory endurance training leads to an improvement in pulmonary muscle function through its effect on relatively conserved respiratory muscle fibers by increasing capillary and mitochondrial density and overall oxidative enzyme capacity

(Dimarco et al, 1985; Park et al, 2010). Therefore, all subjects who participated in playing harmonica showed improvement caused by respiratory muscle pressure.

After 5-month training, no significant difference was found in PCF. These findings did not support our research hypothesis. Contrary to the findings of our study, a previous study reported that IMT was effective in increasing muscle power and coughing ability (Kang, 2006b). PCF accompanies increases in abdominal and intrathoracic pressures and requires the appropriate strength of the core muscles, and diaphragm and intercostal muscles, which are indispensable when inducing a reflexive cough (Torres et al, 2014). In this regard, a possible reason behind these findings could be that the subjects in this study had a disease that made it difficult to improve such muscles, and harmonica training did not involve similar motions that occur during a cough. Thus, manually assisted techniques were more useful in patients with NMD with PCF level <160 L/min (Torres et al, 2014).

Contrary to our hypothesis, no significant MVV was observed between periods. The possible explanation could be that the subjects who participated in our study used motorized wheelchair for ambulation. Kang et al. (1998) reported that subjects with NMD who use wheelchairs showed significantly lower MVV during IMT compared with the NMD subjects capable of walking. As MVV is a dynamic evaluation indicator, the NMD subjects capable of walking preserved respiratory muscle better than patients with NMD who uses wheelchair. Furthermore, the results were attributable to the characteristics of the neuromuscular disease, in which complete and accurate performance of MVV is difficult, requiring repeated MIP and MEP for 12 seconds as an indicator of the ability to respond when MVV is needed.

This study has several limitations. First, the subjects had different ages and diagnoses. This is because of the difficulty in finding and recruiting NMD patients who could move independently while playing the harmonica as subjects. Subjects with the same diagnosis should be recruited in future studies. Second, this study included a relatively small number of subjects, and training duration and frequency were not individually customized. Therefore, further studies, including large sample size, are necessary to confirm our results. Future studies should determine the long-term effects of playing harmonica on different pulmonary function parameters.

Conclusion

The purpose of this study was to determine whether 5-month training of harmonica can affect pulmonary function by assessing FVC, PCF, MIP, MEP and MVV in patients with NMD. Both supine and sitting positions of FVC, MIP, and MEP significantly increased after the training. Therefore, playing harmonica can be an effective method for improving pulmonary function in subjects with NMD.

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