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Cystic Fibrosis and Exercise : A Systematic Review

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ABSTRACT

Purpose: The aim of this article is to systematically review the literature that examine exercise programs and evaluate their effect in Cystic Fibrosis patients.

Method: MEDLINE, EMBASE and CINHALL, Biological Abst., Up To Date electronic databases were searched from January 2005 to December 2013.

Results: Only seven exercise interventions met the eligibility criteria for review. They include a very short interval of twice in one hour to a long duration of 12 months. The exercise programs were walk, step test, aerobic, breathing, gaming and breathing. Majority had supervised training and assessment. Interventions carried out in home and in hospital setting.

Conclusions: All kinds of exercises helped in improving the respiratory function. Future research scope is also identified in various unexplored areas.

Keywords: Cystic Fibrosis, Exercise, Physical activity, Aerobic, Systematic review

INTRODUCTION

Cystic fibrosis (CF) is a genetic autosomal recessive disease, associated with decline of pulmonary function, pancreatic insufficiency, and important physical limitation^{(1)&(2)}. CF primarily affects the lung function and nutritional status⁽³⁾. Patients with pulmonary disease caused by CF are known to have reduced exercise tolerance^(4,5). Regular physical activity and exercise are considered important for the well-being of these patients⁽⁶⁾.

Aerobic exercise enhances cardiovascular fitness, enhances airway clearance, slows the decline in lung function⁽⁷⁾ and improves body mass.

Regular self-selected physical activity has been shown to improve lung function in adults with CF⁽⁸⁾. Studies demonstrated that time spent in moderate-to-vigorous physical activity correlated to aerobic capacity, independent of lung disease or function in adolescents and adults with CF⁽⁹⁾. Studies have demonstrated the benefits of exercise for CF patients^(10, 11).

Enzyme replacement capsules address digestive problems. Participation in exercise and physical activity are important part of the treatment process⁽¹⁰⁾.

Exercise is recommended for adults with CF to assist with airway clearance and reduce cardiovascular risk factors associated with inactivity⁽¹²⁾. Exercise also associated with an QoL for adults with CF⁽¹³⁾. It has been stated that aerobic capacity correlates with QoL measures and changes in the former are associated with changes in the latter⁽⁸⁾.

Exercise training may benefit children with CF by improving aerobic fitness and increasing muscle mass. Some studies have shown benefits from exercise and physical activity such as increased cardiorespiratory fitness and exercise tolerance⁽¹⁴⁾. Higher levels of aerobic fitness associated with prolonged survival. Investigations into components of exercise programs, which can increase their fitness are highly relevant for patients with CF⁽¹⁴⁾.

Exercises at home is a best method of getting cooperation and acceptance from children⁽¹⁵⁾. A

randomized cross over trial with concealed allocation on gaming console exercise for CF patients were explored⁽¹⁶⁾.

The aim of this article is to systematically review the literature that examine exercise programs and evaluate their effect in CF patients.

METHODS

Search Strategy

We performed a systematic review using the methodology addressed by Cochrane Collaboration Protocol⁽¹⁷⁾. MEDLINE, EMBASE and CINAHL, Biological Abst., Up To Date electronic databases were searched from January 2005 to December 2013. The search terms used were Cystic Fibrosis and Exercise or Physical activity or Aerobic Strengthening. We also searched reference lists from relevant articles and searched journals to identify articles to include in the review. Most recent search was performed on 29th January 2013.

Study Criteria

Articles were included if they meet the following criteria: Participants were adolescent or adults with CF, Study published in English or translation available in English.

Exclusion criteria

Unpublished work and articles, articles which are not published in English language or the translation was not available in English.

Study Selection

The literature search was done by single author many sittings. Duplicates were removed; titles abstracts and full articles were made in the hard copy form and screened repeatedly to fit into the eligibility criteria.

RESULTS

Studies Retrieved

Details of the literature search are provided in Figure 1. The search criteria resulted in retrieval of 678

article, with the Boolean /Phrase cystic fibrosis, Fibrosis and Exercise or Physical activity or Aerobic Strengthening. In the review 624 Academic Journals, 43 Magazines 3 news 2 Trade publications and 2 Dissertations were retrieved. It was decided to include only Academic Journals and removed duplicates. Manual screening was done and retrieved only articles which had an intervention. The reviewers found only 7 articles meeting the criteria.

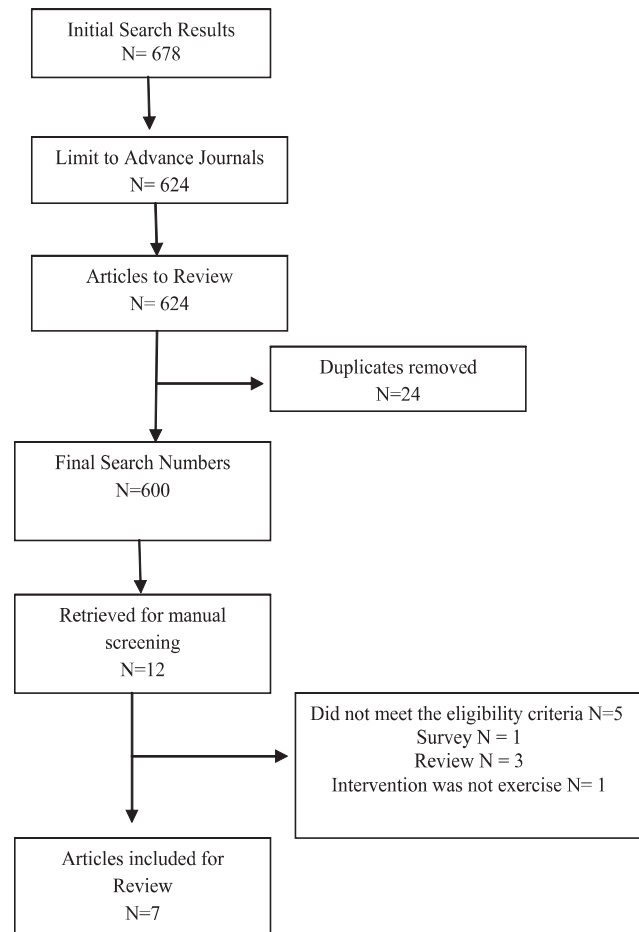


Fig. 1. Flow chart of search results

Study Quality Assessment

Table 1 details the major criteria for methodological study quality assessment. Among 7 none met the full Delphi list criteria⁽¹⁸⁾. An extra criteria of drop out was added because 4 studies reported. Compliance and repeatability of the program was mentioned in all studies. Method of randomization is very clear in all reviewed studies. Two types of interventions mentioned in three studies.

Table 1: Methodological Quality Assessment

STUDY	Holland, A. E., Rasekaba, T., Wilson, J. W., & Button, B. M. (2011).	Ziegler, B., Rovedder, P. M., Oliveira, C. L., Silva, F. e., & Dalcin, P. R. (2010).	Elbasan, B., Tunali, N., Duzgun, I., & Ozcelik, U. (2012).	Schmidt, A. M., Jacobsen, U., Bregnballe, V., Olesen, H. V., Ingemann-Hansen, T., Thastum, M., & OlufSchjotz, P. (2011).	Keochkerian, D., Chlif, M., Delanaud, S., Gauthier, R., Maingourd, Y., & Ahmaidi, S. (2008).	Kuys, S. S., Hall, K., Peasey, M., Wood, M., Cobb, R., & Bell, S. C. (2011).	Paranjape, S. M., Barnes, L. A., Carson, K. A., Loosen, H., & Mogayzel, P. J., Jr. (2012).
	Desaturation During the 3-Minute Step Test Predicts Impaired 12-Month Outcomes in Adult Patients With Cystic Fibrosis	Repeatability of the 6-Minute Walk Test in Adolescents and Adults With Cystic Fibrosis	Effects of chest physiotherapy and aerobic exercise training on physical fitness in young children with cystic fibrosis	Exercise and quality of life in patients with cystic fibrosis: A 12-week intervention study	Breathing Pattern Adopted by Children with Cystic Fibrosis with Mild to Moderate Pulmonary Impairment during Exercise	Gaming console exercise and cycle or treadmill exercise provide similar cardiovascular demand in adults with cystic fibrosis: a randomised cross-over trial	Exercise improves lung function and habitual activity in children with cystic fibrosis
A method of randomization performed	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Treatment allocation was concealed	Yes	Yes	Yes	No	No	Yes	Unclear
Groups similar at baseline regarding the most important prognostic indicators			Yes		Yes	Yes	
Eligibility criteria specified	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Outcome assessor blind	Unclear	Unclear	Yes	No	No	Yes	
Compliance rate reported	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Drop-out reported	Yes	Yes	Yes	Yes	No	No	Yes
Exercise sessions supervised	Yes	Yes by a Physiotherapist	Yes (Physiotherapist and pediatric pulmonologist)	No	Yes (skilled pediatric pulmonary function technicians who regularly work with children with CF)	Yes (Physiotherapist)	Unclear

Cohort Details

Studies were examined for the number of participants, type of study. The study characteristics are presented in Table II. The sample size ranged from 18 to 101. A total of 291 subjects participating across the seven studies.

The age across the study participants were mentioned in 6 and one study mentioned the mean age of the participants, which shows that the participants were not different in their age. Clinical stability of participants was mentioned in six studies. One study categorized participants as mild, moderate and severe cases⁽¹⁹⁾.

Exercise Intervention

Exercise interventions were examined for their number of groups in the study, type of intervention, mode, location of intervention, supervised or not, duration of intervention. Table III describing the details of exercise intervention. The intervention for the study groups included step test, walking, aerobic, physiotherapy, gaming and cycling. Assessment time varied from twice in 60 mins to a maximum of 12 months. Six studies interventions were supervised by a skilled personal⁽²⁰⁾. 100% compliance reported in two studies^(16,21). Four studies were having single group and three studies had two groups for their intervention.

Table 2. Participants Details

Study	Group	Sample	Gender		Age	Severity of CF
			M	F		
Holland, A. E., Rasekaba, T., Wilson, J. W., & Button, B. M. (2011).	Step test	101	56	55	18-63	Normal lung fun 23 Mild disease 22 Moderate Disease 34 Severe Disease 22
Ziegler, B., Rovedder, P. M., Oliveira, C. L., Silva, F. e., & Dalcin, P. R. (2010).	Walk	31	12	19	15-49	Clinically stable
Elbasan, B., Tunali, N., Duzgun, I., & Ozcelik, U. (2012).	Physiotherapy Aerobic	20	Not mentioned		13-May	Clinically stable
Schmidt, A. M., Jacobsen, U., Bregnballe, V., Olesen, H. V., Ingemann-Hansen, T., Thastum, M., & Oluf Schiøtz, P. (2011).	Aerobic exercise program	24	Not mentioned		14-50	Not clear
Keochkerian, D., Chlif, M., Delanaud, S., Gauthier, R., Maingourd, Y., & Ahmaidi, S. (2008).	Breathing exercise	9CF 9 Healthy	2 2	7 7	14-Nov	Not hospitalized at least 1 month before procedure
Kuys, S. S., Hall, K., Peasey, M., Wood, M., Cobb, R., & Bell, S. C. (2011).	Gaming console Exercise using either a treadmill or cycle ergometer	19	10	9	28 (mean)	Clinically stable
Paranjape, S. M., Barnes, L. A., Carson, K. A., von Berg, K., Loosen, H., & Mogayzel, P. J., Jr. (2012).	The types of activity were chosen by the subject in consultation with the physical therapist and included a wide variety of sports, play, and physical extracurricular activities.	78	45	33	6-16	Severe lung disease and growth failure

Table 3: Details of Exercise Intervention

Study	Groups	Intervention Type	Mode	Intervention Location	Frequency	Duration	Adherence (Calculated from drop outs)	Intervention Duration
Holland, A. E., etal. (2011).	One group	Stepstest	3 Minute Step Test	Hospital outpatient clinic	During the clinic visit	3 Minute 15cm step	96%	12 months
Ziegler, B., etal. (2010).	One group	Walk	6 Min walk in a 30m corridor	Hospital	2 times, 6 min walk in 60 min gap	6 min walk in a 30m corridor repeat after 60 mins	87.1%	Twice in 60 mins
Elbasan, B., etal. (2012).	Two group	Physiotherapy Aerobic Exercise	Breathing Techniques Treadmill	Hospital	Chest measurement in 3 different times Physiotherapy 6 weeks duration	Sit up test 20m distance in 20m shuttle run test 10 step 15 cm step stair climbing test	80%	6 weeks
Schmidt, A. M., etal.(2011).	One group	Aerobic exercise	12 week individually tailored and unsupervised aerobic exercise	Hospital CF center	Measurements taken at the start and end of the study	Instructed to exercise half an hour three times a week for 12 weeks Warm up 5 mins 25 mins of supervised training during the visit	58.3%	12 weeks

Table 3: Details of Exercise Intervention (Contd.)

Study	Groups	Intervention Type	Mode	Intervention Location	Frequency	Duration	Adherence (Calculated from drop outs)	Intervention Duration
Keochkerian, D., etal. (2008).	Two group	Breathing exercise	Subjects underwent firstspirometric measurements at rest. Then sat on the electromagnetic braked cycle ergometer (ER 900, Jaeger) to perform a continuous incremental cycling protocol. Workload were individualized for each patient based on clinical factors to provide exhaustion between 8 and 10 min of exercise.	Not clear	Only once on the same day	1 st Spirometric measurement at rest Electromagnetic braked cycle ergometer (ER 900, Jaeger) to perform a continuous incremental cycling protocol	100%	48 hr period within 72 hrs of discharge
Kuys, S. S., etal.. (2011).	Two group	Experimental • Gaming console 15 min • Usual care Control • Cycle ergometry or treadmill walking and running 15 min • Usual care	Intervention • Gaming console Exercise with treadmill or cycle ergometer	Physiotherapy Gym of the Adult Cystic Fibrosis Unit (Hospital)	The two exercise interventions were conducted for all participants within a 48 hour period, within 72 hours of discharge.	25 mins 10 mins rest and 15 mins exercise And a rest period excluded in the calculation	100%	Within 48 hours 2 interventions
Paranjape, S. M.,etal.(2012).		Activity were chosen by the subject in consultation with the physical therapist included a wide variety of sports, play, physical extracurricular activities.	Two-month exercise regimen with selected activities by the subject and designed with the clinic physical therapist	Exercise at home	20-30 minutes of moderate to vigorous activity at least five times a week	20 -30 mins mild to moderate activity 5 times a week	75.6%	2 months

OUTCOMES

Outcomes of the intervention programs are reported. All studies are reported in two categories of measures: pulmonary function and fitness outcomes.

FEV₁ % predicted was examined in all seven studies. BMI was assessed in six studies. One study checked only the weight. Table 1V showing the outcomes of the studies.

Table 4: Summary of Outcomes

Study	Activity		Mean ±SD	Range
Holland, A. E., Rasekaba, T., Wilson, J. W., & Button, B. M. (2011).	3 Minutes Step Terst	FEV1 Predicted FVC % predicted FEV1/FVC (%) BMI (Kg/m ²) Resting SpO ₂ (%)	61+23 80+20 63 + 14 22.2 + 4.2 95 _ 2	23-122 14-131 37-95 14.1-51.3 89-100
Ziegler, B., Rovedder, P. M., Oliveira, C. L., Silva, F. e., & Dalcin, P. R. (2010).	Walk	BMI (kg/m ²) FEV1 (% predicted) FVC (% predicted)	20.8+ 2.2 61+ 28 71+ 23	16.4-26.1 15-119 30-130
Elbasan, B., Tunali, N., Duzgun, I., & Ozcelik, U. (2012).	Physiotherapy Aerobic Exercise	FVC FEV1 (%) %FEF25-75 (%)	89.37 ± 13.81 87.15 ± 13.12 87.25 ± 14.89	72-107 80-109 61-112

Table 4: Summary of Outcomes (Contd.)

Study	Activity		Mean \pm SD	Range
Schmidt, A. M., Jacobsen, U., Bregnballe, V., Olesen, H. V., Ingemann-Hansen, T., Thastum, M., & Oluf Schiøtz, P. (2011).	Aerobic exercise program	FEV1% n =13 (*) BMI z score n =14	87.6 (74.8;105.8) -0.34 (-0.87;0.43 -0.41	85.0 (76.5;99.0) (-0.92;0.49)
Keochkerian, D., Chlif, M., Delanaud, S., Gauthier, R., Maingourd, Y., & Ahmaidi, S. (2008).	Breathing exercise	BMI FEV1, % predicted FVC, % predicted FEV1/FVC, % RV/TLC, %	Control 21.1+1.5 107+6 103+3 85+4 22+3	CF 20.6+1.6 66+7 79+3 69+6 32.9+8.9
Kuys, S. S., Hall, K., Peasey, M., Wood, M., Cobb, R., & Bell, S. C. (2011).	Intervention • Gaming console Exercise using either a treadmill or cycle ergometer	FEV1 (% pred), FVC (% pred),	51 (21) 71 (16)	
Paranjape, S. M., Barnes, L. A., Carson, K. A., von Berg, K., Loosen, H., & Mogayzel, P. J., Jr. (2012).	Exercise regimen consisted of activities chosen by the subject	BMI FEV1, % predicted	56 99	2-96 32-132

DISCUSSION

It was an effort to find a best good way of exercise for patients with CF. It is a known fact that exercise helps in CF. The search found that there were very less studies conducted in this field. From 2005 to 2012 the reviewers could find only seven studies which are having an exercise intervention for CF. In this review it was found that the intervention varied from 2 times in one hour to 12 months, settings home and hospital and interventions were supervised and non supervised. Although the studies were conducted in different setting with various kinds of intervention and duration and supervision are different in no case intervention or exercise gave a negative effect in respiratory function or in the BMI. The 3 minute step test⁽¹⁹⁾ reported as a good way to improve the lung function and decrease hospital stay at 12 months.

Exercise Intervention Designs

The exercise interventions were clearly described and documented for the repeatability in majority of the studies. Only one study had the intervention on the same day in 60 minutes rest period after the first assessment⁽²²⁾. One study was conducted as individually tailored unsupervised home based aerobic exercise program⁽²⁰⁾. This study reported the maximum drop outs (58.3%). One factor for the drop out may be lack of supervision in exercise. Several factors may influence adherence to any treatment regime, including a patient's willingness to participate

in regular exercise over a period of time. Enjoyment and perceived competence in an activity or exercise have been suggested to be the most important⁽²³⁾. All other studies reported a compliance above 70%. Short duration of exercise was examined in one study⁽¹⁶⁾. All exercise programmes irrespective of its kind improved the lung function of the subjects. Strength training for Cystic Fibrosis patients is still a novel area for further study.

Location

Of the seven exercise programmes 5 of them conducted in the hospital one all exercise sessions were done in home setting. All the hospital based exercise sessions reported a greater compliance among the participants. The main issue of home based aerobic exercise program was it was unsupervised. The lung function was reported high when the training sessions were supervised. Four studies conducted as single group^(15, 19, 20, 22) that means there was no control group for the intervention.

Intervention Duration

The reviewed studies varied in their intervention duration from twice in one hour interval to 12 months. In one study the training and the assessment took place in an hour time⁽²²⁾. The longest duration was 12 months in which participants were assessed on their each visit to the clinic⁽¹⁹⁾. The short term interventions showed a fast change in the respiratory function.

FUTURE RESEARCH

There is very limited studies to investigate the exercise intervention in CF patients. There is a good future to identify a best exercise programme for CF. Combination of exercise interventions will attract the attention of the clients. Life experiences, QOL and impact of exercise programme⁽²⁰⁾ of CF patients can be explored in a future study. Clinical outcomes for 3 min step test and 6 min walk exercise can be compared^(19, 22). Nutritional status and exercise tolerance of CF is another area to be explored⁽¹⁴⁾. Longer duration and large number of samples can be included as a future plan^(15, 16).

CONCLUSION

Regular exercise is very important in maintaining good health. This is very important in patients with chronic illness. Exercise is considered as an important tool in CF care. All the health team members involved should be encouraging CF patients to carry out exercise programs regularly and there should be an assessment for the improvement of the lung function which will motivate the patients. Future research should focus on intervention with control groups and larger samples. Real life experiences of Cystic fibrosis patients can be examined.

Ethical Consideration: Authors used published articles for the review purpose.

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