Aerobic exercise improves lung function in children with intellectual disability: a randomised trial

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Question: In children with intellectual disability, is lung function lower than in healthy peers and does it improve with exercise? **Design**: Randomised trial with intention-to-treat analysis and assessor blinding. **Participants**: Forty-four 12-year old children with Down syndrome or other intellectual disability with an average IQ of 42 (SD 8). **Intervention**: The experimental group performed aerobic exercise for 30 minutes, five days per week, for eight weeks. The exercise was supervised walking, running, and cycling, with a target of moderate intensity. The control group continued usual activities and performed no specific exercise. **Outcome measures**: Lung function as FEV₁ and FVC in litres was measured with spirometry at baseline and after the intervention at eight weeks. Prior to the baseline measures, all participants underwent familiarisation of spirometry for one week. **Results**: At baseline, FEV₁ of the children with intellectual disability was a mean of 87% (95% CI 83 to 91) and FVC was 94% (95% CI 91 to 97) of predicted normal values. After intervention, FEV₁ had increased by 160 ml (95% CI 30 to 290) and FVC by 330 ml (95% CI 200 to 460) more in the experimental group than the control group. **Conclusion**: An 8-week program of aerobic exercise improves lung function in children with intellectual disability significantly. **Trial registration**: ACTRN12609000365268. **[Khalili MA, Elkins MR (2009) Aerobic exercise improves lung function in children with intellectual disability: a randomised trial. Australian Journal of Physiotherapy 55: 171–175]**

Key words: Intellectual disability, Lung function tests, Down syndrome, Aerobic exercise

Introduction

Respiratory infections are common in young people with Down syndrome and some other forms of intellectual disability (Baird and Sadovnik 1988, Friez et al 2006), accounting for a large proportion of the early mortality seen in these groups (Chaney and Eyman 2000, Yang et al 2002). Various predisposing factors to respiratory infections have been identified. These include swallowing difficulties, reflux, and other co-morbidities in people with intellectual disability (Fryers 1984, Turner and Moss 1996), and poor sinus drainage, congenital airway strictures, and immunological abnormalities in people with Down syndrome (Saenz 1999, Nespoli et al 1993, Yang et al 2002, Schloo et al 1991, Shapiro et al 2000). Even when well, children and adolescents with Down syndrome or other forms of intellectual disability appear to have reduced lung function compared to healthy, age-matched controls (Dichter et al 1993, Pastore et al 2000). This may limit their ability to withstand the above insults to the lungs. However, the studies of lung function in these groups have contradictory results and are limited by small cohorts or substantial missing data. That data are lost is due largely to inadequate technical performance of spirometry by the participants (Pastore et al 2000), potentially skewing the results. With substantial practice, however, children and adolescents with Down syndrome are able to achieve reproducible and valid results on spirometric testing (Dichter et al 1993). Further valid data are required.

One factor that could contribute to poor lung function is reduced abdominal muscle performance. Field tests consistently demonstrate that abdominal strength and endurance are reduced in children and adolescents with intellectual disability (Corder 1966, Rarick et al 1970, Londeree and Johnson 1974, Pizarro 1990). The reduction may be extreme, with mean values below the first percentile of the results of healthy, age-matched controls (Dichter et al 1993). Adolescents and children with intellectual disability typically do less vigorous activity and recreational activity than their peers (Sharav and Bowman 1992, Whitt-Glover et al 2006). Low activity levels are therefore a likely contributor to poor abdominal muscle strength and endurance.

As with many other patient populations (Taylor et al 2007), aerobic exercise appears to have worthwhile benefits in children and adolescents with intellectual disability. Ozmen and colleagues (2007) conducted a randomised trial of a 10-week aerobic exercise regimen in 8-15 year olds with intellectual disability, demonstrating a significant improvement in exercise capacity on the 20 m Shuttle Run Test. Millar and colleagues (1993) conducted a similar study in older adolescents and young adults with Down syndrome. Although physiological measures of cardiovascular fitness did not improve, significant gains were made in peak exercise time and grade on an incremental treadmill test, indicating improved exercise capacity. Aerobic exercise may also improve lung function but to our knowledge this has not been examined in a randomised trial in children with intellectual disability. The research questions therefore

- 1. Compared to data from healthy children, do children with intellectual disability have reduced lung function when they are well and after they have had substantial practice with the spirometry test?
- 2. What is the effect of aerobic exercise on lung function in children with intellectual disability?

Method

Design

A randomised trial with intention-to-treat analysis and assessor blinding was conducted. Participants were recruited from children who attended day rehabilitation at the Shafa Rehabilitation Centre in Semnan, Iran. After their eligibility was confirmed, participants were familiarised with the spirometry procedure for one week before baseline measurements were conducted. Following this, participants were randomly allocated to one of two groups by flipping a coin. Eligibility was therefore determined a week before group allocation was randomly determined, thus allowing concealed allocation. The experimental group undertook eight weeks of aerobic exercise while the control group carried out their usual activities. Outcomes were measured at baseline and after the 8-week intervention by a physiotherapist with more than five years of clinical experience who was blinded to group allocation throughout the study. Participants and the staff supervising the exercise sessions were not blinded to group allocation.

Participants

Children were eligible for inclusion if they had intellectual disability, as indicated by their diagnosis in their medical record and by their IQ score, and if the attending physician, in conjunction with the occupational therapist, determined that they could co-operate with the assessment and exercise procedures and that they could undertake exercise safely. They were excluded if they had major motor, behavioural, cardiovascular, or respiratory co-morbidities, or if they were acutely unwell. At baseline, age, height, and gender were recorded to allow the calculation of lung function values predicted by normative equations. Body weight was recorded to allow calculation of body mass index. IQ was recorded from the medical notes.

Intervention

The experimental group undertook 30-minute, supervised, exercise sessions, five times per week for eight weeks. The exercise modalities were walking, running, and cycling. Walking and running were carried out on flat ground and were followed by cycling on a cycle ergometer. Participants performed 10 minutes of each modality at each session with no break between modalities. The target intensity was moderate, determined by the sports coach who asked the participants about their level of exertion, noted that the respiratory rate was elevated but that they could still participate in conversation, and regularly palpated pulse rate at the wrist. They also conducted their usual daily activities (eg, self care, and group activities such as printing, painting, hand craft, and theatre), but no other specific exercises were carried out during the period of the study.

The control group conducted only their usual daily activities and no other specific exercises.

Outcome measures

The primary outcome was the forced expiratory volume in one second (FEV₁) in litres. The secondary outcome was the forced vital capacity (FVC) in litres. Spirometry was performed and analysed according to the American Thoracic Society/European Respiratory Society guidelines (American Thoracic Society/European Respiratory Society 2005). Participants ate only a light meal at their last meal before testing. All tests were conducted at an ambient

Table 1. Baseline characteristics of participants, therapists and centres.

Characteristic	Participants randomised (n = 44)		
	Exp (n = 24)	Con (n = 20)	
Age (yr), mean (SD)	11.9 (1.5)	11.5 (1.6)	
Height (m), mean (SD)	1.58 (0.06)	1.58 (0.05)	
BMI (kg/m²), mean (SD)	18 (2)	18 (2)	
Gender, n males (%)	13 (54)	12 (60)	
Intellectual disability, n (%)			
Down Syndrome	21 (87)	17 (85)	
Other	3 (12)	3 (15)	
IQ (points), mean (SD)	43 (7)	41 (9)	

Exp = experimental group, Con = control group

temperature of 19 degrees Celsius and humidity of 23%.

Participants were familiarised with spirometric testing for one week before baseline measurements were conducted. All participants attended five 25-minute sessions during the week prior to randomisation. At these sessions, a physiotherapist with five years experience supervised practice of the spirometric procedure. Participants received demonstrations of the correct procedure and verbal feedback about their technique. Participants were able to perform the procedure correctly and reliably (American Thoracic Society/European Respiratory Society 2005), demonstrating less than 10% variation between the final two FVC values before commencing the intervention.

Data analysis

In the absence of an established minimum clinically-important difference in FEV_1 in this population, we nominated 0.30 litres. The best estimate of the standard deviation of FEV_1 in a population of children with Down syndrome after training of their spirometric technique is 0.32 l (Dichter et al 1993). A total of 38 patients would provide an 80% probability of detecting a difference of 0.30 l in FEV_1 at a two-sided 5% significance level. To allow for some loss to follow-up, we increased the total sample size to 44.

Baseline characteristics are presented using descriptive statistics. Baseline lung function values were converted to percentages of the normal predicted value for each participant's age, gender, and height according to the equations of Quanjer and colleagues (1995). Independent t-tests (95% CI) were used to compare the between-group difference in FEV₁ and FVC from baseline to Week 8.

Results

Flow of participants, therapists and centres through the trial

Forty-four participants were recruited and underwent familiarisation and baseline testing. Randomisation allocated 24 to the experimental group and 20 to the control group. The baseline characteristics of the two groups are presented in Table 1 and in the first two columns of Table 2. All participants completed the intervention as allocated and

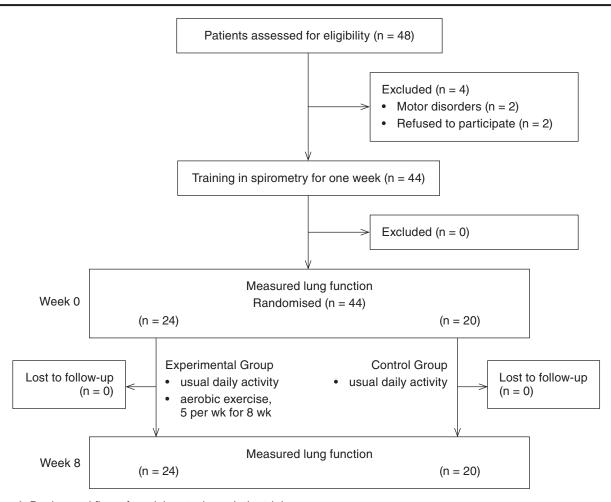


Figure 1. Design and flow of participants through the trial.

all completed post-intervention measurement at 8 weeks (Figure 1).

Exercise sessions were supervised by a sports coach who had eight years of experience including three years working specifically with people with disabilities. Outcomes were measured by two physiotherapists who each had at least five years' experience, including three years working specifically with people with disabilities.

Only one centre, the Shafa Rehabilitation Centre of Semnan, Iran, was involved in the study. This centre provides day rehabilitation to 60 children. All are aged 14 years or younger, all have intellectual disability, and only outpatient teaching and rehabilitation services are offered. Down syndrome accounts for around 85% of the caseload.

Compliance with trial method

In preparation for the baseline measures, all participants attended the familiarisation prior to baseline testing. All participants in the exercise group attended all of their 40 scheduled exercise sessions. No participants in the control group attended any of the exercise sessions.

Lung function in children with intellectual disability

Normal values for FEV₁ and FVC were derived from data on 5861 healthy children and adolescents, and the standard deviation for both was 11% (Quanjer et al 1995). Overall, the participants of the present study had lower lung function

than predicted by their age, height, and gender. Their mean FEV_1 was 87% (95% CI 83 to 91) of predicted normal, ie, 13% lower than predicted FEV_1 (95% CI 10 to 16). Their mean FVC was 94% (95% CI 91 to 97) of predicted normal, ie, 6% lower than predicted FVC (95% CI 3 to 9).

Effect of intervention

Group data for all outcomes at baseline and Week 8 for experimental and control groups are presented in Table 2 while individual data are presented in Table 3 (see eAddenda for Table 3). Both FEV_1 and FVC improved significantly more in the experimental group than in the control group. After intervention, FEV_1 had increased by 160 ml (95% CI 30 to 290) more in the experimental group than the control group, which is a relative increase of 7% (95% CI 2 to 12). FVC had increased by 330 ml (95% CI 200 to 460) more in the experimental group than the control group, which is a relative increase of 11% (95% CI 6 to 14).

Discussion

The first part of this study identified a statistically-significant reduction in lung function in children with intellectual disability. This was despite the fact that the participants were well and had undergone extensive familiarisation with spirometry. Given the hypothesis that reduced lung function may predispose this population to respiratory infections, this result has implications for further research. The first implication is to investigate whether intervention can improve lung function in this population. The next is

Table 2. Mean (SD) lung function for each group, mean (SD) difference within groups, and mean (95%CI) difference between groups.

	Groups			Difference within groups		Difference between groups	
Lung function	Week 0		We	ek 8	Week 8 minus Week 0		Week 8 minus Week 0
	Exp (n = 24)	Con (n = 20)	Exp (n = 24)	Con (n = 20)	Ехр	Con	Exp minus Con
FEV1 (1)	2.41	2.40	2.58	2.41	0.17	0.01	0.16
	(0.33)	(0.33)	(0.36)	(0.34)	(0.28)	(0.03)	(0.03 to 0.29)
FEV1 (%pred)	86	87	92	87	6	0	6
	(9)	(10)	(11)	(12)	(10)	(1)	(1 to 11)
FVC (I)	3.07	2.96	3.35	2.91	0.28	-0.05	0.33
	(0.38)	(0.34)	(0.44)	(0.37)	(0.21)	(0.23)	(0.20 to 0.46)
FVC (%pred)	95	93	103	91	9	-1	10
	(10)	(12)	(12)	(13)	(7)	(7)	(6 to 14)

Exp = Experimental group, Con = Control group, FEV1 = forced expiratory volume in one second, FVC = forced vital capacity, %pred = % of predicted value for age, height, and gender (Quanjer et al 1995).

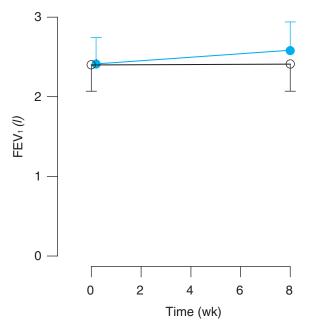


Figure 2. Mean (SD) FEV₁ in the experimental group (closed circles) and the control group (open circles) at baseline and 8 weeks.

to investigate whether intervention found to be effective at improving lung function subsequently reduces respiratory infection.

In the second part of this study, we identified a small but statistically-significant effect of exercise on two measures of lung function: FEV_1 and FVC. The confidence interval around the estimate of the effect on FEV_1 excluded 0.30 l which was the value we nominated as a minimum clinically-worthwhile difference. However, we feel that this should not cause further investigation of exercise as a potential preventive tool against respiratory infections in this population to be abandoned, for several reasons. The first is that, in the absence of data to guide us, the figure of 0.30 l was chosen somewhat arbitrarily. In relative terms, this minimum clinically important difference equates to a 12.5% increase in FEV_1 . In other patient populations, interventions with smaller relative effects on lung function (some even

smaller than our estimate of 7%) are nevertheless effective at reducing respiratory infections (Fuchs et al 1994, Elkins et al 2006). Finally, the effect of exercise on lung function may be able to be increased, perhaps by changing the mode, intensity, or duration of the training. For example, if one of the mechanisms of improvement in lung function as a result of exercise in this population is via improved abdominal muscle strength, then a program with a greater emphasis on abdominal exercises may produce a greater benefit in lung function.

The results are unlikely to have been skewed by an unrepresentative sample, since only two of the 46 eligible children who were invited to participate refused to do so. The high rate of willingness to participate in both parts of the study and the 100% adherence with the exercise sessions and follow-up measurements confirms that the measurement and intervention were well accepted in this population.

It is reassuring that although there were only two outcomes measured in this study, both showed statistically-significant beneficial effects. The effect on FVC was more substantial than that for FEV_1 , both in absolute and in relative terms. Although measuring only lung function minimised the risk of Type I error, this small number of outcome measures can also be seen as a limitation of the study. Measurement of abdominal strength would have helped determine whether it is part of the mechanism by which exercise improves lung function in this group. Measurement of respiratory infections during the 8-week period would have helped with sample size calculation for future research. Another limitation of the study was that therapists and patients were not blinded.

In conclusion, exercise had a small but statistically-significant effect on lung function in children with intellectual disability. However, further research is required to determine whether this is clinically worthwhile or could be increased by modifying the training regimen.

eAddenda: Table 3 available at AJP.physiotherapy.asn.au

Ethics: The ethics committee of the Shafa Rehabilitation Centre approved the study. Written informed consent was obtained from the parents of potential participants before data collection began.

Competing interests: None declared.

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