

Original Article

Levels of moderate–vigorous physical activity are low in Spanish children with cystic fibrosis: A comparison with healthy controls



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Abstract

Background: Physical activity (PA) is recommended as part of the treatment regimen for cystic fibrosis (CF) although objective methods have been scarcely used to monitor achievement of PA guidelines.

Methods: PA was measured by accelerometer in outpatient CF children (n = 47) and results were compared with those obtained in age- and gender-matched healthy controls (n = 39).

Results: 2.1% of the outpatients fulfilled PA guidelines (i.e. ≥ 60 min·day⁻¹ of moderate-to-vigorous PA (MVPA)) vs. 34.2% of controls. Overall, lower MVPA levels were observed in CF patients than controls despite the former undergoing more light or total PA. Peak oxygen uptake was also lower in the CF group than in controls (37.5 ± 7.8 vs. 43.5 ± 7.6 ml·kg⁻¹·min⁻¹, $p = 0.002$) and was correlated with MVPA and vigorous PA in the former.

Conclusions: These findings support a need to promote PA interventions (including MVPA) targeted at improving cardiorespiratory fitness in CF children.

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Keywords: Physical activity; Guidelines; Cardiorespiratory fitness

1. Introduction

Cystic fibrosis (CF) is an inherited genetic disorder that leads to progressive deterioration of lung function and physical capacity; it affects approximately one in 2500 newborns [1]. The median life expectancy of patients with CF is around

31 years [2]. Regular physical activity (PA) has become increasingly important and widely accepted as part of the therapy and rehabilitation programs in CF management [3]; although, PA remains underutilized and not always incorporated into routine CF management [3]. Several studies have reported beneficial effects of exercise training that include: slower lung function decline [4], enhanced airway clearance [5], improved nutritional status and quality of life [6] and reduced hospital admissions [7]. The additional training benefits for CF patients do not differ from those for healthy individuals and include mainly improved aerobic fitness and muscle strength [8,9].

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It is known that there is correlation between PA levels and aerobic fitness (as determined by peak oxygen uptake, VO_{2peak}) [10] and higher aerobic fitness is a predictor of survival in this disease [11–15]. It is consequently of medical interest to quantify actual PA levels in children with CF to help prescribe PA programs designed to improve their aerobic fitness.

Current guidelines indicate that children and adolescents should engage in at least 60 min of moderate to vigorous PA (MVPA) daily [16]. However, given that many children fail to reach this level of activity [17], there is a need to ensure that youths are meeting the PA guidelines recommended for optimal health outcomes. This is especially important in CF children; however, despite the known impact of PA on the health and quality of life of these children, there has been relatively little research evaluating the PA levels of this patient group. One of the reasons for this deficiency is that assessing PA in children using self- or parent-administered questionnaires has many limitations; children find it especially difficult to describe or quantify their PA levels [18]. Such limitations have led to an increasing interest in the use of more objective methods to monitor PA. As opposed to questionnaires, accelerometers can provide minute-by-minute recordings of PA and can therefore be used to objectively quantify PA [19].

To date, there are studies in which PA levels in children [6,20] or adults [21] with CF have been measured objectively by accelerometers. Yet the studies in CF children have some methodological limitations, i.e. use of activity counts rather than activity cut-point thresholds to quantify activity intensity [6] or insufficient number of accelerometer wearing days for appropriate PA analysis [20]. This situation makes it difficult to determine if children were performing light, MVPA or vigorous PA. There is a need for longer periods of monitoring PA objectively and the use of intensity cut-points to obtain information on usual PA levels in children with CF (including the comparison of weekend vs. weekday activity behavior) and on how well these patients comply with current PA guidelines. Such data would add valuable information to the existing literature and help health carers recommend PA more effectively.

The purpose of our study was to objectively measure PA levels in outpatient children with CF vs. age and gender-matched healthy controls over a week (as well as on week days vs. weekends), and to determine the proportion of them meeting the PA guidelines (primary outcome). VO_{2peak} was also determined in the two groups to examine its possible correlation with PA (secondary outcome). We hypothesized that children undergoing treatment for CF would show overall reduced MVPA levels compared to their healthy peers, with the majority of the former not meeting the PA guidelines.

2. Materials and methods

2.1. Participants

The research project was in accordance with the Declaration of Helsinki Research of 1974 (last modified in 2008) and was approved by the corresponding Review Board (Children's

Hospital *Niño Jesús*; Madrid, Spain). Informed consent was obtained from the parents of each participant.

Participants were recruited by screening the medical records of this hospital's Pneumology department to select 113 children under treatment for CF. After the corresponding pneumologist provided consent, subjects were enrolled in the study if they met each of the inclusion criteria: outpatient diagnosed using a genetic test for CF and treated at the aforementioned hospital, and boy/girl aged 6–17 years living in the Madrid region (so that they could attend VO_{2peak} testing sessions). Exclusion criteria were: having severe lung deterioration (forced expiratory volume (FEV_1) < 50% the expected value), an unstable clinical condition (i.e. hospitalization within the previous 3 months), *Burkholderia cepacia* infection, or any condition (e.g. muscle–skeletal disorder) impairing exercise testing. According to these criteria, 73 children with CF were originally enrolled in the study; valid accelerometry data was obtained in 47 (see Fig. 1 for the flow diagram of the participants in the CF group) of whom 39 had also undergone a cardiorespiratory fitness test (see below).

The data of the CF children were compared to those of 39 age and gender-matched controls with complete accelerometry and VO_{2peak} data (see below) pooled from a total cohort of 118 children living in the same neighborhood. The same measurements (using the same methodology and equipment) were performed in the two groups during years 2010–2012, as explained below. Anthropometric and cardiorespiratory fitness evaluations were performed in the exercise physiology laboratory of the Children's Hospital *Niño Jesús* (CF group) and of the European University of Madrid (control group).

2.2. Measurements

2.2.1. Anthropometric data

Height without shoes was measured using a clinical stadiometer (Asimed T2, Barcelona, Spain) and recorded to the nearest millimeter. Body mass was determined to the nearest 0.05 kg using a balance scale (Ano Sayol S.L., Barcelona, Spain) with subjects in their underwear. Body mass index (BMI) was calculated as weight/height ($kg \cdot m^{-2}$). Skinfold thickness was measured with a Harpenden caliper (Holtan Crymych, United Kingdom) at the biceps, triceps, sub-scapular, abdominal, supra-iliac, thigh and calf areas following the criteria described elsewhere [22]. Percentage of body fat [23] was calculated from body density values using age- and gender-specific equations, i.e. for boys and girls aged ≤ 11 years [24], 12–15 years [25], and 16–18 years [26].

2.2.2. Accelerometry

PA was planned to be measured on 7 consecutive days (Thursday to Wednesday or Friday to Thursday) using the Actigraph GT3X accelerometer (Actigraph, Pensacola, FL, USA). All children wore the accelerometer in an elastic waistband on their right hip during the day, except while bathing or undertaking other water activities. Verbal and written instructions for the care and placement of the monitor were given to both the children and their parents. The GT3X is

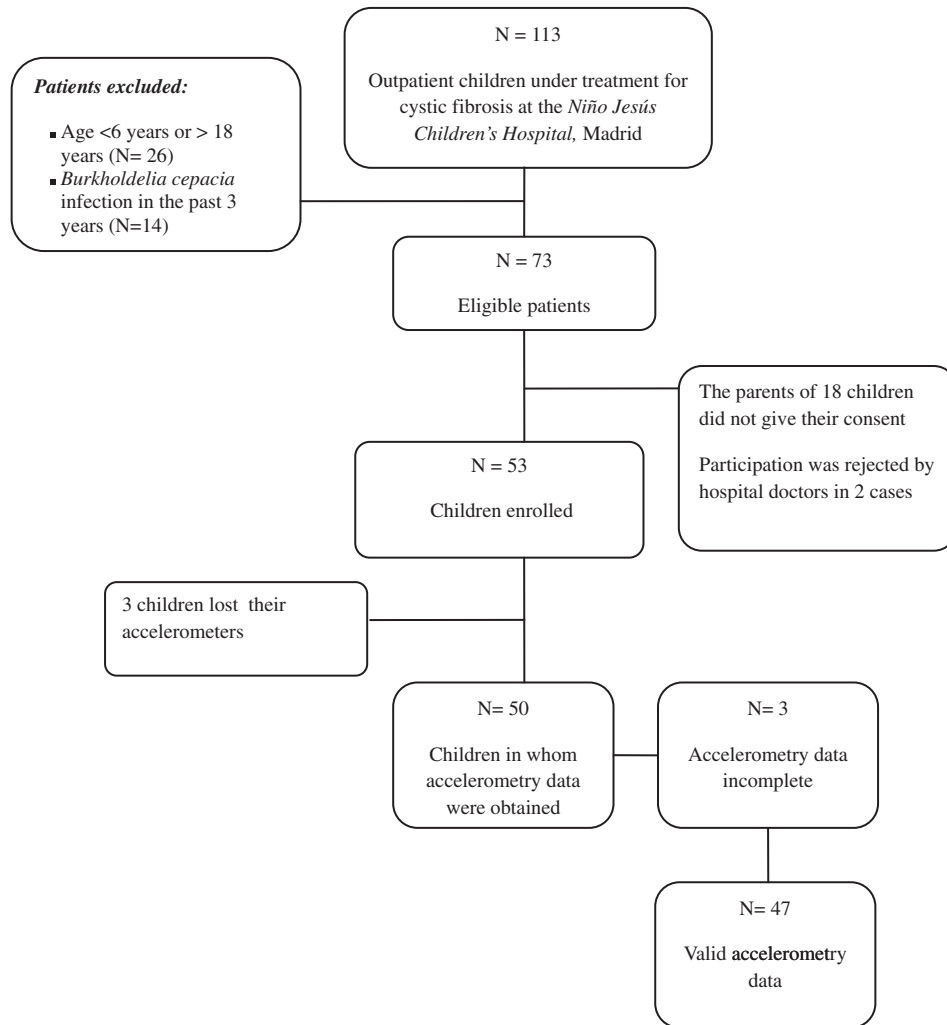


Fig. 1. Flow diagram of the patient recruitment process.

a lightweight (27 g), small ($4.5 \times 3.5 \times 1.0$ cm), tri-axial accelerometer. Movement in the three axes is detected as a combined function of the frequency and intensity of the movement, while an electronic filter rejects motion outside the range of normal human movement. This accelerometer has proven valid and reliable for the determination of PA in children [27].

For each participant, a minimum of 4 days' monitoring including one weekend day, and a minimum of 10 h of complete accelerometry data per day, were considered necessary for PA assessment to be considered valid and thus for their accelerometry data to be included in the study [28]. Epoch duration or the sampling period was set at 15 s, and we applied cut-points (see below) to the 15-second epochs (after dividing by four); output was expressed as counts per minute ($\text{counts} \cdot \text{min}^{-1}$). Following recent recommendations for accelerometry research conducted in children and adolescents [29], count ranges for the various activity intensities were determined using the cut-points originally developed by Evenson et al. [30]: 0 to 100 $\text{counts} \cdot \text{min}^{-1}$ for sedentary activity, 101 to 2295 $\text{counts} \cdot \text{min}^{-1}$ for light PA, 2296 to 4011 $\text{counts} \cdot \text{min}^{-1}$ for moderate PA and >4012 $\text{counts} \cdot \text{min}^{-1}$ for vigorous PA. To analyze the

accelerometer data, Kinesoft software, developed specifically for the Actical and Actigraph accelerometers, was used. The outcome variables were expressed in time ($\text{min} \cdot \text{day}^{-1}$) spent for the different PA-intensity categories. Thus, we calculated how many minutes of light, and moderate and vigorous activity were performed by the children during the full week, as well as on week and weekend days. We excluded from the analysis bouts of 20 continuous minutes of activity with intensity counts of 0, considering these periods to be non-wearing time [28].

2.2.3. $VO_{2\text{peak}}$

Children performed a graded exercise test on a treadmill (Technogym Run Race 1400HC; Gambettola, Italy) to determine $VO_{2\text{peak}}$. Before the test, they undertook at least one practice session. All the exercise tests were conducted under similar environmental conditions (temperature of 20 to 24 °C, relative humidity of 45 to 55%). Gas-exchange data were measured *breath-by-breath* by open circuit spirometry using pediatric face-masks (V_{max} 29C, SensorMedics; Yorba Linda, CA, USA). Peak oxygen uptake ($VO_{2\text{peak}}$) was recorded as the highest mean VO_2 value obtained for any continuous 20-second period. Heart rate (HR) was continuously monitored during the tests from a

Table 1
Anthropometric and cardiorespiratory data by group.

	CF group		Control group		<i>p</i> -Value
	No. with complete data	Result	No. with complete data	Result	
<i>Anthropometric variables</i>					
Weight (kg)	47	40.1(11.7)	39	45.1(12.3)	0.07
Height (cm)	47	146.9(15.8)	39	150.4(14.8)	0.30
BMI (kg·m ⁻²)	47	18.6(2.8)	39	19.9(2.1)	0.07
∑ skinfolds (mm)	47	77.5(31.5)	39	85.5(32.4)	0.31
Fat mass (%)	47	19.8(5.9)	39	21.8(6.0)	0.17
Fat free mass (%)	47	80.2(5.8)	39	78.2(6.0)	0.17
<i>Cardiorespiratory fitness</i>					
VO _{2peak} (ml·kg ⁻¹ ·min ⁻¹)	39	37.5(7.8)	39	43.5(7.6)	0.002

Data expressed as mean(SD). Abbreviations: BMI, body mass index; CF, cystic fibrosis; VO_{2peak}, peak oxygen uptake. Significant between-group differences appear in bold.

12-lead ECG. Treadmill speed was started at 1.5 km·h⁻¹ (for the smaller children, height <120 cm) or 2.5 km·h⁻¹ (if children's height was >120 cm) at a 0.5% gradient. Thereafter, both treadmill speed and gradient were increased (by 0.1 km·h⁻¹ and 0.5% respectively) every 15 s. The tests were terminated upon volitional fatigue, when the child was unable to keep up the required workload, or if the child suffered a coughing fit [23]. All the children were verbally encouraged and had no visual access to their parents.

2.3. Statistical analysis

Data are provided as mean ± standard deviation (SD). Tests of normality, symmetry and kurtosis were performed. Between-group differences in PA data were assessed using independent Student's *t*-tests. Chi-square tests were used to examine differences between the two groups in terms of fulfilling PA guidelines. Correlations between PA variables and VO_{2peak} were examined by ordinary least squares (OLS) linear regression. All statistical tests were performed using the PASW software (v. 19.0 for WINDOWS, Chicago). Significance was set at *p* < 0.05.

3. Results

The main descriptive data for the study groups are provided in Table 1. A full set of accelerometry data was obtained in 47 CF children (24 boys, 23 girls, mean age: 12 ± 3 years). Of these, VO_{2peak} was recorded in 39 (23 boys). The control group (n = 39) included 23 boys and 16 girls aged 12 ± 2 years. The children's anthropometric data did not differ between the two groups (all *p* > 0.1). Mean VO_{2peak} was significantly higher in the control group than in the CF group (*p* = 0.002).

PA data are shown in Table 2. No significant difference was found between the two groups in minutes of total accelerometry time registered per day. Sedentary time was significantly higher for the control group when all week, week days and weekend days were considered (all *p* = <0.001). Total PA and light PA levels for the whole week were significantly higher (*p* = 0.001 and *p* = 0.040 respectively) in patients than controls. However, weekly vigorous PA and MVPA were significantly lower in the

former (*p* = 0.001 and *p* = 0.020 respectively). Significant differences were also detected when separated analysis were done for week days and weekend days. Thus, during week days, the CF group undertook more total (*p* < 0.001) and light PA than the control group (*p* = 0.001), but spent less sedentary time (*p* < 0.001), as well as vigorous PA (*p* < 0.001). Similar differences between the two groups were found during weekend days.

Overall, 34.2% (n = 13) vs. 2.1% (n = 1) of children in the control and CF group respectively met the MVPA guidelines (*p* < 0.001) (Fig. 2). The percentages of children fulfilling MVPA guidelines during week (*p* < 0.001) or weekend days (*p* = 0.04) were also lower in the CF group.

In the CF group, VO_{2peak} was significantly correlated with all-week MVPA (r = 0.46, *p* = 0.003) and all-week vigorous

Table 2
Physical activity (PA) data by group.

	CF group (n = 47)	Control group (n = 39)	<i>p</i> -Value
Average monitoring time (min·day ⁻¹)	725(74)	764(97)	0.249
<i>All week</i>			
Sedentary time (min·day ⁻¹)	362(67)	484(85)	<0.001
Total PA (min·day ⁻¹)	363(85)	280(58)	0.001
Light PA (min·day ⁻¹)	319(71)	226(44)	0.040
MVPA (min·day ⁻¹)	44(28)	54(15)	0.020
Vigorous PA (min·day ⁻¹)	9(6)	17(9)	0.001
<i>Week days</i>			
Sedentary time (min·day ⁻¹)	407(73)	300(101)	<0.001
Total PA (min·day ⁻¹)	361(90)	283(35)	<0.001
Light PA (min·day ⁻¹)	315(63)	226(44)	0.001
MVPA (min·day ⁻¹)	46(33)	57(19)	0.60
Vigorous PA (min·day ⁻¹)	8(9)	17(9)	<0.001
<i>Weekend days</i>			
Sedentary time (min·day ⁻¹)	355(79)	463(83)	<0.001
Total PA (min·day ⁻¹)	355(96)	275(79)	<0.001
Light PA (min·day ⁻¹)	319(77)	230(62)	<0.001
MVPA (min·day ⁻¹)	36(32)	45(20)	0.164
Vigorous PA (min·day ⁻¹)	8(9)	16(6)	0.01

Data expressed as mean(SD). Abbreviations: CF, cystic fibrosis; MVPA, moderate-to-vigorous physical activity; PA, physical activity. Significant between-group differences appear in bold.

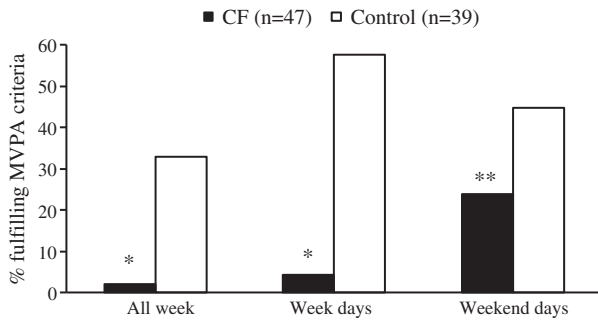


Fig. 2. Percentages of children meeting recommendations for moderate-to-vigorous physical activity (MVPA) by group. * $p < 0.001$, ** $p = 0.04$ for between-group comparisons. Abbreviation: CF, cystic fibrosis.

PA ($r = 0.39$, $p < 0.05$). When analyzing data on week and weekend days, VO_{2peak} was also significantly correlated with MVPA ($r = 0.41$, $p = 0.009$ and $r = 0.39$, $p < 0.05$ respectively) and vigorous PA ($r = 0.38$, $p < 0.05$ and $r = 0.35$, $p < 0.05$ respectively). No significant correlations between PA and VO_{2peak} were found in the control group except for vigorous PA on weekend days ($r = 0.35$; $p < 0.05$).

4. Discussion

The main finding of our study was that, overall, the children with CF spent less time undertaking intense PA compared to the age-matched controls; moreover, children with CF had a lower achievement of PA guidelines. Compared to controls, CF group spent significantly more time in light PA and total PA but less time in MVPA and vigorous PA. Current PA guidelines recommend that children engage in at least 60 min and up to several hours of MVPA on all or most days of the week [16]. Only one child with CF fulfilled this PA guideline during the whole week and a very small proportion of the patients (4.3%) met the guideline on week days. A larger proportion (23.9%) met the guideline during weekends. Among the control children ~59% fulfilled the PA guideline on week days and ~34% met the guideline during all week. When considering the total amount of light PA and total activity performed, the key difference emerging between the groups was the *intensity* rather than the *quantity* of PA performed over a full week. In other words, the children with CF engaged in a reasonable *quantity* of PA (i.e. total PA) but this PA was of insufficient *quality* (i.e. MVPA and vigorous PA) to meet general PA recommendations.

The PA results of this study did not agree, at least apparently, with those of Kilbrige et al. (2012) [20] and Selvadurai et al. (2004) [6], where patients with CF were as active and fit as their healthy controls. With regard to this, there is evidence [31] that a minimum of 7-day monitoring is needed to provide a reliable estimate of usual PA behavior in children and adolescents and to assess differences in weekend vs. week day activity behavior, yet the study by Kilbrige et al. [20] only used 3 days of monitoring PA. Moreover, the use of counts and a global measure of PA instead of the amount of minutes of light, moderate or vigorous PA could have altered the results of

previous research [6], together with the use of different types of accelerometer among studies. RT3 accelerometers used in Kilbride et al. [20] were compared to the Actigraph accelerometers and SenseWear Armband (SWA) in free-living energy expenditure (EE) within sedentary environments [32] and the RT3 model was found to be less valid for quantifying EE during light-intensity activities than Actigraph accelerometers and SWA. Recently, Rabinovich and colleagues [33] have recommended the use of GT3X Actigraph accelerometer as the most valid monitor to measure daily life PA in patients with chronic obstructive pulmonary disease.

Our results are in agreement with those of Nixon et al. (2001) [34] where children with CF engaged in less vigorous PA than their healthy non-CF peers, despite having good lung function. However, comparisons between studies are difficult because they used a questionnaire (i.e. Kriska's Modifiable Activity Questionnaire) to estimate PA. Similar results have been found in young adults [21] where subjects wore a SWA during 5–7 days, and they reported that the amount of PA above moderate intensity was significantly reduced in CF compared with controls, which could imply that the differences found in our study may be still present in young adulthood.

Fulfillment of the above-mentioned PA guideline in Spanish children [17] has been separately analyzed for 9- and 15-year-old children: the guideline was met in 60% of the boys and 34.1% of the girls (9-year-olds) and in 28.8% of the boys and 8.9% of the girls (15-year-olds). This same study reported a higher rate of guideline fulfillment during week days versus weekend days. The control children examined here showed a similar pattern. In contrast, our CF patients were better able to fulfill the PA guideline during weekends. Parents' awareness of the need that their CF children engage in vigorous PA might explain, at least partly, higher fulfillment of guidelines during weekends, when parents have more time to share with their children and can have a more proactive role in outdoor activities and sports playing.

VO_{2peak} was significantly correlated with all-week, week-day and weekend day MVPA and vigorous PA in our CF group. Good aerobic fitness (as determined by VO_{2peak}) is a predictor of survival in CF [12] such that this correlation highlights a need to promote higher intensities of PA. Research has demonstrated the benefits of aerobic and strength exercise training programs for health and quality of life. However, the CF patient has to face challenges and even barriers to participate in PA programs. Mean VO_{2peak} levels are usually lower in patients with CF compared with their healthy counterparts [34]. This variable is an important predictor of mortality in these patients [12,14]. Thus, mortality is high in children with VO_{2peak} values of $< 32 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ (60% after 8 years) whereas it is null in those with VO_{2peak} levels of $> 45 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ [12,14]. In our study, 23.1% ($n = 9$) of the children with CF had a VO_{2peak} lower than $32 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$; 59% ($n = 23$) of patients had a VO_{2peak} between 32 and $45 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$, and only in 17.9% ($n = 7$) was this indicator higher than $45 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$. These data support the notion that PA interventions (including vigorous PA) aimed at improving cardiorespiratory fitness in child CF patients are urgently needed, at least in Spanish CF patients.

In conclusion, we observed that only 2.1% of children undergoing treatment for CF achieved PA guidelines. Overall, compared to controls they performed a greater quantity of PA (i.e. minutes of total PA) but this PA was of insufficient intensity. These findings prompt a need to encourage children with this illness to participate in sports, games and activities that involve bouts of vigorous PA from an early age.

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