Is Measuring Physical Literacy in School-Aged Children With Cystic Fibrosis or Congenital Heart Disease Needed?

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Purpose: To explore the association between cardiorespiratory fitness and other physical literacy domains in children with cystic fibrosis (CF) or congenital heart disease (CHD).
Methods: In 28 children with CF (n = 10) or CHD (n = 18), aged 7 to 11 years, cardiorespiratory fitness and the following physical literacy domains were measured: (*a*) physical competence, (*b*) motivation and confidence, (*c*) knowledge and understanding, and (*d*) daily behavior (ie, self-perceived moderate-to-vigorous physical activity [MVPA]).
Results: Cardiorespiratory fitness was significantly associated with motivation and confidence and self-perceived MVPA. There were no other significant associations.
Conclusions: Cardiorespiratory fitness is associated with self-perceived MVPA, motivation, and confidence in children with CF or CHD. (Pediatr Phys Ther 2023;35:43–47)
Key words: cardiorespiratory fitness, congenital heart disease, cystic fibrosis, physical activity, physical literacy

INTRODUCTION

Physical activity is essential for children's physical and mental well-being[1] and considered vital to enhance cardiorespiratory fitness (CRF) in children with a chronic medical condition (CMC) such as cystic fibrosis (CF) and congenital heart disease (CHD).[2,3] Although CF and CHD differ in terms of pathophysiology, treatment, and trajectory, in both conditions, CRF is associated with health status and future mortality.[4,5] CF is an inherited genetic disorder that affects the gastrointestinal and reproductive tracts but leads primarily to progressive deterioration of lung function and CRF. CF is incurable and affects approximately one in every 2500 live births.[6] As a result of medical and therapeutic advances, life expectancy has increased during the last decade to 49 years for those born in 2017.[7] CHD is characterized by structural defects of the heart and occurs in approximately 9 in every 1000 live births.[8] Severity of the disease varies from case to case, but survival in children with CHD has increased because of surgical advances. More than 90% of the children with CHD now reach adulthood.[9]

Physical literacy is a concept that helps to understand why people are physically active or inactive. The concept is described as the physical competence, motivation, confidence, knowledge, and understanding to value and take responsibility for engagement in physical activities for life.[10] Higher levels of physical literacy in healthy children are associated with higher levels of physical activity.[11] Physical literacy has not been studied in children with CF or CHD.

According to the World Health Organization recommendations, healthy children need moderate-to-vigorous physical activity (MVPA) for at least 60 minutes a day, with strength, flexibility, and coordination exercises to improve or maintain CRF at least 3 times a week.[12] Guidelines from the European Cystic Fibrosis Society, the American Heart Association, and the Association for European Pediatric Cardiology advise children with CF or CHD to comply with the recommendations of 60 minutes or more of MVPA, with a few exceptions for children with CHD who have specific lesions or complications.[2,3,13] Although the importance of physical activity is accepted, a number of children do not meet the recommendations for physical activity[14,15] and children with CF or CHD may participate even less in physical activity than their peers.[16,17]

As pediatric physical therapists are often involved in stimulating physical activity to enhance CRF in children with CF or CHD,[18] insight into determinants of physical literacy for CRF provides pediatric physical therapists' tools for more personalized treatment. Therefore, the aim of the current study was to

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explore associations between CRF and the other physical literacy domains in children with CF or CHD.

MATERIALS AND METHODS

Study Sample

Twenty-eight children with CF (n = 10) or CHD (n = 18), aged 7 to 11 years, treated in the Wilhelmina Children's Hospital, University Medical Centre Utrecht, were invited to participate in this cross-sectional observational study between December 2020 and July 2021. Children with insufficient knowledge of the Dutch language were excluded. The Medical Research Ethics Committee of the University Medical Centre Utrecht approved this study (nr: 20-565/C). All parents gave written informed consent for their child's participation. All children gave verbal assent.

Procedure

Children were evaluated directly after a clinical visit to the pediatric physical therapist, whereby measurement of CRF was assessed if requested as part of standard medical care. Assessment, without measurement of CRF, took approximately 15 minutes. Measurement of CRF took approximately 30 minutes.

The theoretical framework of the Canadian Assessment of Physical Literacy-Second Edition (CAPL-2) was used to investigate children's physical literacy.[19] The CAPL-2 distinguishes 4 domains: (*a*) physical competence, (*b*) motivation and confidence, (*c*) knowledge and understanding, and (*d*) daily behavior. Physical competence was assessed with a maximal cardiorespiratory exercise test (CRF) and the Canadian Assessment of Motor Performance (CAMSA) (motor competence). Motivation and confidence, knowledge and understanding, and daily behavior were assessed with a questionnaire. Assessment took place in a quiet room at the hospital. Because the children in the current study were younger than the children in previous studies using this questionnaire, great care was taken to ensure that the children understood the questions. All questions were read aloud by the researcher to all the children.

Physical Competence.

Cardiorespiratory Fitness. CRF, expressed as predicted peak oxygen uptake (L/min) (predicted $\dot{V}O_{2peak}$), is the gold standard for assessing CRF.[20] Children performed a maximal cardiorespiratory exercise test on a cycle ergometer using the Godfrey protocol.[21] During this test, children breathed through a face mask (Hans Rudolph Inc, Kansas City, Missouri) connected to a calibrated gas analysis system (Ergostik, Geratherm; Accuramed, Lummen, Belgium). $\dot{V}O_{2peak}$ was defined by the mean value of the last 30 seconds during the maximal exercise test. Predicted $\dot{V}O_{2peak}$ values (%) were obtained from age- and sex-matched Dutch controls.[22]

Motor Competence. The CAMSA[23] was used to evaluate children's motor competence. Children are instructed to complete a circuit of movement exercises as fast and good as possible performance of the skills. A total score (1-28) is calculated on the basis of time (in seconds) (1-14) and quality of the performance (0-14). The performance of the CAMSA was captured on video, with time and quality of the performance scored from video. Two assessors (J.N. and M.S.) independently scored the quality of the performance. Any disagreement was discussed until agreement was reached. The CAMSA is a valid and reliable measure to investigate motor performance in healthy children aged between 8 and 12 years.[23]

Motivation and Confidence. Motivation and confidence were assessed with 12 items from the CAPL-2 questionnaire.[24] Six items are assessed with a structured alternate-response format, whereby each question consists of 2 contradictory quotes. Children first have to choose which quote describes them best and indicate whether this is either "a little bit true for me" or "totally true for me." The other 6 items are assessed on a 5-point Likert scale. Each of the 12 items is scored with a maximum of 2.5 points such that the maximum score per aspect is 7.5 and the maximum total score for motivation and confidence is 30 points. Internal consistency for motivation and confidence in this study was high (Cronbach $\alpha = 0.94$).

Knowledge and Understanding. Knowledge and understanding were assessed with 5 items from the CAPL-2 questionnaire.[25] This questionnaire records children's knowledge about recommended physical activity guidelines, terminology related to CRF and muscle strength, and how to enhance specific aspects of their physical competence. Four of the 5 questions have a multiple-choice question format and concern: (*a*) daily physical activity guidelines, (*b*) how children improve their sports skills, and definitions of (*c*) CRF and (*d*) muscle strength. The final question consists of a short story about physical competence, whereby children complete 6 missing words. Each correct answer in the knowledge and understanding assessment was assigned with 1 point, resulting in a maximum score of 10 points.

Daily Behavior. The CAPL-2 was used to investigate self-perceived MVPA.[26] Children were asked to specify how many days during the past week they were physically active for at least 60 minutes a day, whereby their heart rate and breathing were increased.

Data Analysis

Descriptive data are presented as means with standard deviations (continuous outcome measures) and numbers and percentages (dichotomous outcome measures). All analyses were performed in SPSS version 25.0 (IBM Corp, Armonk, New York). Level of significance was set at $P < .05$. Data distributions of every continuous outcome measure were checked using Kolmogorov-Smirnov tests, Shapiro-Wilk tests, skewness, kurtosis, and eyeballing. Next, Spearman (nonparametric) or Pearson (parametric) correlations were calculated between all variables. When significant, correlation coefficients were classified as weak ($0.1 \leq r < 0.3$), moderate ($0.3 \leq r < 0.5$), or strong ($r \geq 0.5$).[27]

RESULTS

A total of 28 children (21 boys/7 girls), aged 7.6 (SD = 1.1) years, participated in this study. No adverse effects occurred during the assessment of tests. The total sample consisted of 10 children with CF (6 boys/4 girls), mean age 8.6 (SD = 1.4) years,

and 18 children with CHD (15 boys/3 girls), mean age 7.1 (SD = 0.2) years. Types of CHD were as follows: transposition of the great arteries (n = 10), tetralogy of Fallot (n = 4), single ventricle program (n = 3), and hypoplastic left heart complex (n = 1).

Children with CF were significantly older and had higher weight and length than children with CHD (all *P*s < .05). Knowledge and understanding were also higher in children with CF (all *P*s < .001). No differences were found for predicted $\dot{V}O_{2peak}$ ($P = .73$), motor performance ($P = .09$), motivation and confidence ($P = .41$), and self-perceived MVPA ($P = .75$). Table 1 reports mean scores and standard deviations for the total sample and for the children with CF and CHD separately.

Associations Between Physical Literacy Domains

CRF, expressed as predicted $\dot{V}O_{2peak}$ (%), was significantly, moderately associated with motivation and confidence ($r = 0.44$, $P = .02$) and self-perceived MVPA ($r = 0.41$, $P = .03$). No other significant associations were found (all *P*s > .05) (Table 2).

DISCUSSION

CRF was significantly associated with self-perceived MVPA and motivation and confidence but not with motor performance and knowledge and understanding. The association between CRF and physical activity is in line with previous research and clinical guidelines[2,3,13] and supports the importance of participation in physical activity to enhance CRF in children with CF or CHD. The strength of the association was moderate, which is comparable with previous studies in children with CHD or CF.[28,29] This indicates that other factors, such as motivation and confidence, may also be important to predict CRF.

Pediatric physical therapists often focus on stimulating children's motor performance to increase participation, functional mobility, and (physical) activity levels,[18] but we found no association between motor performance and physical activity or between motor performance and CRF. This result is in line with previous research in children with CHD[29] and indicates that motor performance is not the restraining factor for physical activity in children with CF or CHD. Pediatric physical therapists should therefore focus more on stimulating motivation and confidence to enhance CRF in children with CF or CHD, instead of focusing on children's motor performance. This can be done by incorporating behavioral strategies in interventions, for example, using the self-determination theory.[30] More specifically, the self-determination theory considers 3 basic needs for motivation to enhance someone's motivation. These basic needs for motivation are as follows: (*a*) autonomy, (*b*) competence, and (*c*) relatedness. *Autonomy* refers to the need to experience oneself as initiator and regulator of one's actions, *competence* refers to the perception of one's performance, and *relatedness* refers to experience satisfactory relationships with others (eg, therapist, peers). Various studies reported positive associations between these basic needs for motivation and (intrinsic) motivation.[31,32] We argue therefore that pediatric physical therapists should pay specific attention to children's autonomy, competence, and relatedness to increase children's motivation and confidence in order to enhance CRF. Subsequently, although contradictory to our results, higher levels of (intrinsic) motivation are associated with more physical activity,[33] which was significantly associated with CRF in our study.

TABLE 1

Participant Characteristics[a]

Variables	Total Sample (N = 28)	Children With CF (n = 10)	Children With CHD (*n* = 18)	Sig.
Boy/Girl,	21/7	6/4	15/3	.21[b]
Age, y	7.6 (1.1)	8.6 (1.4)	7.1 (0.2)	<.001[c]
Weight, kg	27.3 (5.1)	30.6 (6.0)	25.4 (3.4)	.02[c]
Height, cm	130.7 (8.2)	136.5 (8.9)	127.5 (5.8)	.01[c]
Physical competence				
Motor performance (1-28)	14.0 (3.7)	15.9 (4.7)	12.9 (2.6)	.09[d]
Predicted $\dot{V}O_{2peak}$, L/min (%)	79.4 (17.7)	77.8 (20.7)	80.3 (16.3)	.73[d]
Motivation and confidence (6.6-30)	24.7 (4.7)	26.0 (3.5)	24.0 (5.2)	.41[c]
Knowledge and understanding (0-10)	5.8 (2.0)	7.4 (1.6)	4.9 (1.5)	<.001[c]
Daily behavior				
Self-perceived participation in MVPA, n				.75[c]
0 d	0	0	0	
1 d	0	0	0	
2 d	3	0	3	
3 d	3	3	0	
4 d	4	1	3	
5 d	5	1	4	
6 d	2	2	0	
7 d	11	3	8	

Abbreviations: CF, cystic fibrosis; CHD, congenital heart disease; MVPA, moderate-to-vigorous physical activity.
[a]Data are presented as mean (SD) or as number.
[b]Fisher's exact test.
[c]Mann-Whitney *U* test.
[d]Independent-samples *t* test.

TABLE 2

Correlations Between Physical Literacy Domains

	Physical Competence—Predicted $\dot{V}O_{2peak}$, L/min (%)	Physical Competence-Motor Performance	Motivation and Confidence	Knowledge and Understanding
Physical competence—predicted $\dot{V}O_{2peak}$, L/min (%) (n = 28)	1.00			
Physical competence—motor performance (n = 28)	− 0.11	1.00		
Motivation and confidence (n = 28)	0.44[a]	0.27	1.00	
Knowledge and understanding (n = 28)	− 0.05	0.31	0.03	1.00
Self-perceived participation in MVPA (n = 28)	0.41[a]	0.16	0.35	− 0.12

Abbreviation: MVPA, moderate-to-vigorous physical activity.
[a]$P < .05$.

No other significant associations were found, which is contradictory to the study by Delisle Nyström et al,[34] in which children without medical issues aged 8 to 12 years were evaluated. A possible explanation for this result is the large sample size in the previous study (n = 5307)[34] compared with the smaller sample size in the present study (n = 28). Moreover, Delisle Nyström et al found only small to moderate associations between physical literacy domains (0.10-0.42).

Limitations

We used the CAPL-2 as a framework to investigate physical literacy in children with CF or CHD, but we did not use all measures as proposed by the CAPL-2. More specifically, we did not objectively investigate children's physical activity (eg, accelerometer or pedometer), while physical activity is considered the main outcome of physical literacy. Children with CHD were 7 years old, while the CAPL-2 is developed for children 8 to 12 years old. Therefore, we were unable to compare results with normative scores.

This is the first study that investigated the measurement of physical literacy in children with a CMC in a hospital setting. We found that the CAPL-2 is quick and easy to administer and used low-cost materials. No negative side effects were reported, and all invited parents and children were willing to participate. This makes the CAPL-2 a feasible tool for pediatric physical therapists to assess physical literacy in children with CF or CHD. We used a cycle ergometer with gas analysis to assess children's CRF, which is considered the gold standard for assessing CRF.[20]

CONCLUSION

Physical literacy is feasible and easy to assess in children with CF or CHD during their 1-day hospital visit using the CAPL-2. This study supports that CRF was significantly associated with physical activity, motivation, and confidence but not with motor performance and knowledge and understanding. Pediatric physical therapists are advised to assess all physical literacy domains instead of motor performance only to get a broader insight into determinants of CRF and physical (in)activity. In doing so, more personalized prescription can be given to enhance CRF and physical activity in children with CMC. More specifically, pediatric physical therapists can modify their intervention based on the physical literacy domain(s) children with CMC experience problems in. Future research should investigate physical literacy in other populations of children with CMC and measure physical activity objectively to get better insight into the associations with the physical literacy domains.

What This Adds to the Evidence

Our study is the first to investigate physical literacy in children with CF or CHD. We found that physical literacy can be assessed safely and easily and that CRF was significantly associated with physical activity, motivation, and confidence. The results of this study call for researchers to investigate physical literacy in other populations of children with CMC. In doing so, pediatric physical therapists can provide more personalized care to stimulate physical activity and improve CRF. In addition, there is a need for a standardized physical literacy test for children younger than 8 years, because low physical activity levels are already present at a younger age. Future research should therefore investigate if physical literacy can be evaluated in younger children and, if so, provide normative scores for this age range.

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