


REVIEW ARTICLE

Physical activity assessments in children with congenital heart disease: A systematic review

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Abstract

Aim: To describe the physical activity (PA) of children with congenital heart disease (CHD) according to different assessment methods and compare their results with the World Health Organization (WHO) recommendations.

Methods: A systematic review was conducted using five databases. We included cross-sectional, longitudinal, observational studies and clinical trials in a paediatric population with CHD. In publications with indirect measurement, the score in each dimension was considered. Similarly, moderate-to-vigorous PA (MVPA) was considered as the main outcome in articles with direct measurement.

Results: A total of 1103 articles were found, and 16 primary articles were considered. Eight articles evaluated PA with indirect methods, six with direct methods and two used both methods, representing 1649 subjects evaluated. It was found that 46% of children with CHD do not exceed WHO recommendations for MVPA, with no differences depending on the severity of CHD.

Conclusion: There are a variety of ways to measure PA in children with CHD. In the articles that objectively evaluated PA, the most measured outcome was the MVPA, which shows that the MVPA time was shorter in about half of the children with CHD than what is recommended by WHO.

KEYWORDS

children, congenital heart defects, moderate-to-vigorous physical activity, physical activity

1 | INTRODUCTION

Congenital heart disease (CHD) corresponds to structural defects in the morphogenesis of the heart or its main vessels.¹ This disease is classified according to severity as follows: simple, moderate or complex.² The prevalence is estimated to range between 2.4 and

13.7/1000 live births, and its incidence is around 8/1000 live births in the United States and Europe.^{3,4}

Despite the increase in the life expectancy of children with CHD,⁵ the residual defects remaining after surgery can have a negative impact on morbidity and mortality.⁶ This has reoriented the paradigm of therapeutic management with the aim to optimise the

Abbreviations: AoS, aortic stenosis; CG, control group; CHD, congenital heart disease; HRQOL, health-related quality of life; MET, metabolic equivalent of task; MVPA, moderate-to-vigorous physical activity; PA, physical activity.

health-related quality of life (HRQOL), which evaluates the cognitive, social, emotional and physical aspects.⁷ In this aspect, physical activity (PA), understood as any body movement that demands an expenditure of energy on the baseline,⁸ requires the integration of multiple organs and systems, so any dysfunction in these will impact the normal development of PA.

It is possible to categorise PA in three levels according to the metabolic equivalent of task (MET) necessary to perform an activity.⁹ Thus, the level of expenditure ranges between 1.1 and 2.9 MET for sedentary PA and between 3.0 and 5.9 MET for moderate PA and is ≥ 6.0 MET for vigorous PA. To achieve health benefits, the World Health Organization (WHO) recommends 60 minutes/day of moderate-to-vigorous PA (MVPA; ≥ 3.0 MET).¹⁰

Although it has been reported that physical capacity, understood as the maximum physical effort that a subject can perform, and PA are determinants of morbidity and mortality in adults with CHD^{8,11,12} and, in children with CHD it is significantly lower in comparison with healthy controls,^{13,14} it has been stated that MVPA is the variable that has a direct relationship with increases in self-reported HRQOL.^{15,16} Several studies have described the impact of CHD on PA in children, reporting the risk of suffering other cardiovascular and metabolic diseases during adolescence^{4,7,17,18} and adulthood.^{8,12}

Our objective was to describe studies that investigated the PA of children with CHD using different assessment methods and compare their results with the WHO recommendations.

2 | METHODS

The search was performed using the following databases: Medline, Cumulative Index to Nursing & Allied Health Literature (CINAHL), EMBASE, ProQuest and Scientific Electronic Library Online (SCIELO). This review was carried out in compliance with Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) guidelines.

Peer-reviewed studies that fulfilled specific Patient, Intervention, Comparison, Outcomes (PICO) modified to meet PCO requirements are Patient/Population: Children and adolescents with congenital heart disease; Comparison: Healthy controls; Outcomes: Physical activity level measured through objective and subjective measures. The search strategy is described in the supplementary material (Table S1).

We selected studies conducted on human beings in children under the age of 18, with a diagnosis of CHD. We selected articles published without language restriction between 01 of January 1950 and 31 of March 2020. The following designs were considered: case series, cross-sectional studies, longitudinal observational studies and clinical trials. We excluded articles with duplicate results; articles that group data with adult patients and/or another disease in which the analysis by subgroup was performed.

Potentially eligible items were reviewed based on the article's title and its summary. Then, we read the text extensively to verify its suitability for final inclusion. Two researchers (RAD and RTC)

Key notes

- A wide variety of methods are used to measure physical activity in children with congenital heart disease.
- A significant number of children with congenital heart disease have a shorter moderate-to-vigorous physical activity time than is recommended by the World Health Organization.
- A sedentary lifestyle is common in this population and could harm health and quality of life.

independently extracted the data from the selected articles and recorded them in an ad hoc spreadsheet of relevant data. This included author, country, year of publication, sample size, study design, the age of the subjects, diagnoses, evaluation instruments, evaluated variables and results. The differences obtained from the data extraction were resolved through consensus. In the case of not reaching an agreement, a third researcher (JRF) resolved the differences.

In articles with measurements obtained through direct methods, we extracted the arithmetic or median average results related to the use of accelerometers for a minimum of 3 days or indirect calorimetry, daily PA level with the number of steps per day, sedentary time in minutes and/or inactivity, time in minutes of PA in any of its categories (mild, moderate, vigorous) and/or basal and total daily energy consumption. In articles using indirect measurements, we extracted data for the score obtained in each dimension evaluated by PA self-report questionnaires and/or parent report or recorded in the children's diaries. In articles where no results were expressed with values of methodological relevance, an email was written to the author at the correspondence address requesting the information.

The methodological quality in each article was evaluated by two independent researchers (JRF and RTC) through the verification of compliance with the items of the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) initiative.¹⁹ This instrument was constructed to establish the sections that should be considered in the report of a study with an observational design. If there were differences in the score assigned for the articles, it was decided that they would be resolved by consensus. If no agreement was reached, a third investigator (IRN) resolved the differences.

3 | RESULTS

We found 1103 articles from the five databases. The systematic search sequence and the reasons for excluding articles are presented in Figure 1. After following the search strategy and agreeing on the selection, a total of 16 articles were considered,²⁰⁻³⁵ representing a total of 1649 subjects.

All the articles were written in English. Eight articles used indirect methods for measuring PA,²⁰⁻²⁷ six reported direct methods³⁰⁻³⁵ and

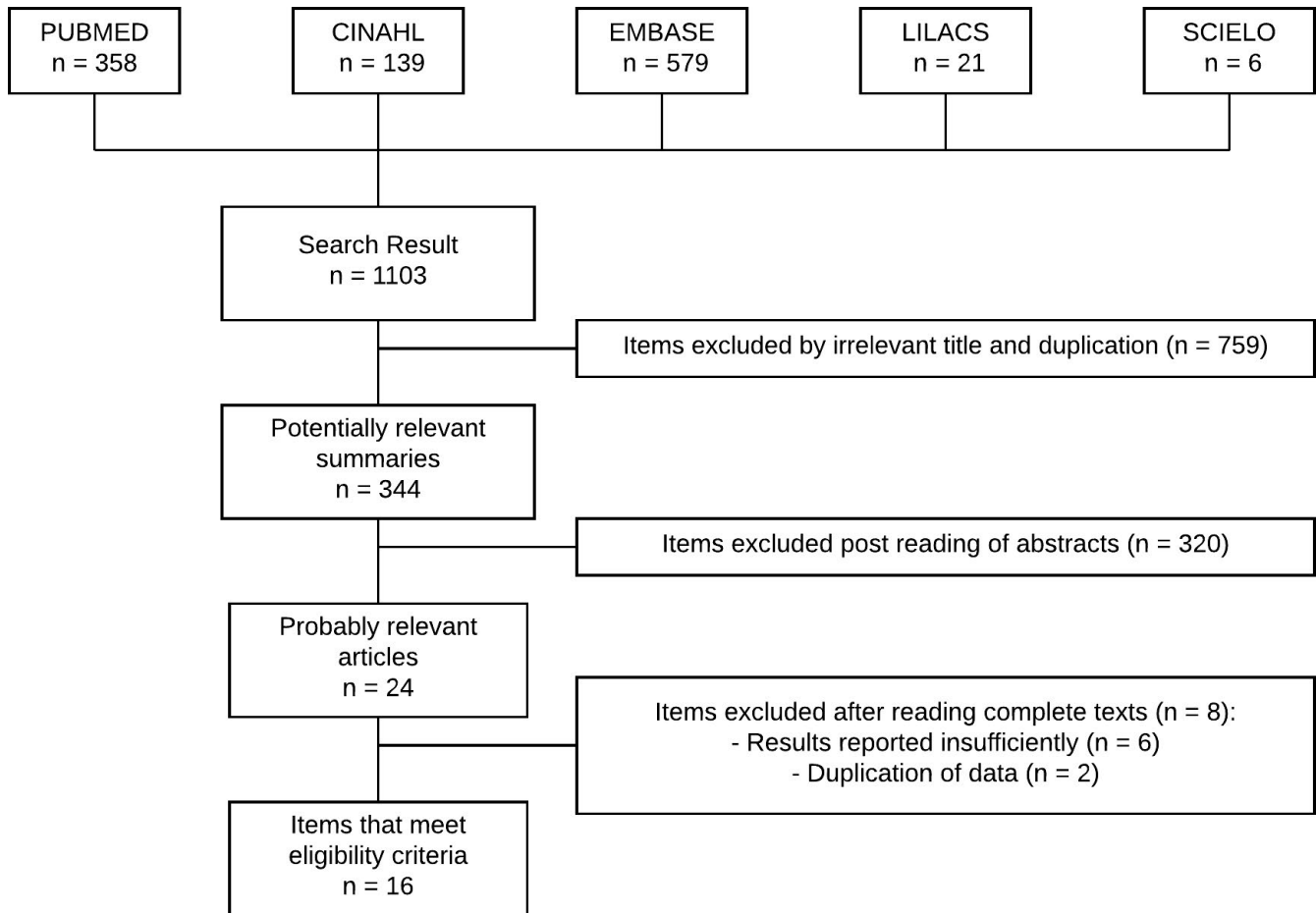


FIGURE 1 Systematic search sequence

two used both.^{28,29} The years of publication ranged between 1994 and 2019; the sample size ranged between 7 and 258 subjects.

All the articles fulfilled between 12 and 21 items of the STROBE initiative. The median compliance was 18 (81.8%; Table 1).

3.1 | Indirect PA measurement

Articles with indirect PA measurement methods reported using self-report surveys that parents completed about their children's PA, direct application surveys to children and self-report surveys completed by children (Table 2).

Using a self-report questionnaire for parents, Casey et al²⁰ reported on an overview of the activity of children with CHD in which 100% of the parents considered that their children have at least moderate limitations when participating in sports.

Reybrouck et al²¹ used a questionnaire with a response scale from 0 to 8 points to describe the level of PA in children, and they classified them under each type of heart defect comparing it with a control group (CG). In two measurements separated by three years, that study found a significantly lower value in patients with Aortic Stenosis (AoS), Tetralogy of Fallot (TOF) and patients with Fontan

surgery (Fontan-PO) in comparison with the CG, of which the Fontan-PO group reported the lowest values.

Bar-Mor et al²² used a self-report survey with 12 possible consensual activities that children could perform. That study used a Likert scale to establish the level of restriction of PA imposed by the cardiologist (1-4), the family (1-4), the PA that the child felt capable of performing (self-efficacy) (0-100) and the extent to which the child participated in each PA (1-5). In the latter, no gender differences were observed. There was a significant correlation between restrictions imposed by the cardiologist vs family and between self-efficacy and the level of PA. The association between the severity and PA level was poor.

Lunt et al²³ employed a self-report survey for children that detailed their participation in organised games and non-organised PA during school time, in summer and in winter. Grouping the data (winter and summer), 71% of the children between the ages of 12 and 14 engaged in adequate or vigorous PA, while 65% of the children between the ages of 15 and 18 did not. Among those who dropped out of school, 54% were classified as inactive. Grouping by sex, 73% of the boys with CHD declared themselves to be active in summer and 62% were active in winter, versus 82% and 74% of the CG, respectively. There was a significant difference in the groups; the PA

TABLE 1 Summary of the evaluation of the methodological quality of primary articles (STROBE)

Author	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	Total
Casey (1994)	1	1	1	0	0	1	0	1	0	0	1	0	1	1	1	0	0	1	0	1	0	1	12/22 (54.5%)
Reybroeck (1995)	1	1	1	1	1	1	1	1	0	0	1	1	1	0	1	0	0	1	1	1	0	1	16/22 (72.7%)
Bar-Mor (2000)	1	1	1	1	1	1	1	1	0	0	1	0	0	0	1	0	1	1	0	0	0	0	12/22 (54.5%)
Leitch (2000)	1	1	1	0	1	1	1	1	0	0	1	0	0	1	1	1	1	1	1	1	0	1	16/22 (72.7%)
Lunt (2003)	1	1	1	1	1	1	1	1	1	0	1	1	1	1	1	1	1	1	1	1	1	0	20/22 (90.9%)
Arvidsson (2009)	0	1	0	1	1	1	1	1	0	1	1	1	1	1	1	1	1	1	1	1	0	1	18/22 (81.8%)
Kao (2009)	1	1	1	1	1	1	1	1	0	0	1	1	0	1	1	0	1	1	1	1	1	1	19/22 (86.4%)
Pasquali (2009)	1	1	1	1	1	1	1	1	0	0	1	1	1	1	1	1	0	1	1	1	1	0	18/22 (81.8%)
Ray (2011)	1	1	1	1	1	1	1	1	0	1	1	1	1	1	1	1	0	1	1	1	1	0	19/22 (86.4%)
Ewalt (2012)	0	1	1	0	1	1	1	1	0	0	1	1	1	1	1	0	1	1	1	1	1	1	17/22 (77.3%)
Chen (2012)	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	0	1	1	1	1	1	21/22 (95.5%)
Stone (2015)	0	1	1	0	1	1	1	1	0	1	1	1	0	0	1	1	0	1	1	1	1	1	16/22 (72.7%)
Banks (2017)	1	1	1	1	0	1	1	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	20/22 (90.9%)
Voss (2017)	1	1	1	1	1	1	1	1	0	0	1	1	1	1	1	1	1	1	1	1	1	1	20/22 (90.9%)
Brudy (2020)	0	1	0	1	1	1	0	1	0	0	1	1	1	0	1	1	1	1	0	1	0	0	13/22 (59.1%)
Moschovi (2019)	1	1	1	1	1	1	1	1	1	1	1	1	0	1	1	0	1	1	1	1	1	0	19/22 (86.4%)

Note: 1: Title and abstract; Introduction: 2: Background/Rationale; 3: Objectives; Methods: 4: Study design; 5: Setting; 6: Participants; 7: Variables; 8: Data sources/Measurement; 9: Bias; 10: Study size; 11: Quantitative variables; 12: Statistical methods; Results: 13: Participants; 14: Descriptive data; 15: Outcome data; 16: Main results; 17: Other analyses; Discussion: 18: Key results; 19: Limitations; 20: Interpretation; 21: Generalisability; Other information: 22: Funding of the study.

TABLE 2 Summary table of primary articles that only incorporate indirect measures of physical activity

Author (y)	Country	n (CHD/ non-CHD)	Study design	Age (y)	CHD DIAGNOSIS CATEGORISED	Assessment	Outcome	Results
Casey et al, 1994 ²⁰	Canada	52 (26/26)	CSS-CG	8.78 ± 3.3	Class III ^a (n = 26)	Questionnaire of Bowyer	PA perceived by parents: School, walking, stairs, running, sport, tiredness. Scale 0-24	70% walking <100 yards; 15% never running 15% never do sports
Reybrouck et al, 1995 ²¹	Belgium	79 (without non-CHD)	LS-CG	9.1 ± 3.2	Class I ^a (n = 27); Class II (n = 40); Class III (n = 12)	Questionnaire of Weymans	Activities: type and number of hours of sports participation, leisure time, participation in physical education at school, transfer to and from school. Scale 0 to 8	Low value of PA in patients with Class II and III compared to healthy controls.
Bar-Mor et al, 2000 ²²	Israel	100 (without non-CHD)	CSS	15.21 ± 1.89 (range 12-18)	Shunt injuries ^b (n = 32); Obstructive injuries (n = 36); Cyanotic injuries (n = 26); Others (n = 4)	Questionnaire of physical activity	Twelve common PA. Scale between 1 and 5.	PA measured score: 2.8 ± 0.7 (range 1.5-4.3); Correlation between cardiologist and family restriction: r = .84 (P < .01); Correlation between Self-efficacy and PA level: r = .50 (P < .001); Association between severity and PA level: r = .11 (P = ns)
Lunt et al, 2003 ²³	Australia	153 (95/58)	CSS- PNAD	14.6 ± 1.8 (range 12-18)	Moderate CHD ^c (n = 110); Severe CHD (n = 43)	New South Wales (NSW) Schools Fitness and Physical Activity Survey	Habitual physical activity levels, psychological determinants and advice received.	Active participation CHD vs PNAD (boys and girls): Winter: 62% vs 74%; Summer: 73% vs 82% (P = .027); Vigorously active children CHD vs PNAD: Winter: 48% vs 67% (P < .02), Summer: 48% vs 69% (P = .04); Similar, but not significant, trends were found when comparing girls surveyed with healthy peers
Pasquali et al, 2009 ²⁴	United States	106 (without non-CHD)	CSS-CG	Median 14.2 (range 6.0-19.9)	Class II or III ^a	Modified Youth/ Adolescent Activity questionnaire	Number of hours/weeks of total, vigorous, and sedentary activity	Level of PA in NW vs OW + OB (h/wk): ST: 40.5 (15.0-95.0) vs 40.0 (15.0- 125.0), P = ns; TPA: 11.5 (0.8-52.3) vs 9.8 (1.7-41.2), P = ns; VPA: 2.5 (0-31.9) vs 4.5 (0-14.9), P = ns. Restriction in any activity (Cardiologist vs Parent): NW: 27% vs 42%, P = ns; OW + OB: 27% vs 38%, P = ns. Restriction in aerobic activity (cardiologist vs parent): NW: 8% vs 26%, P = .04; OW + OB: 6% vs 33%, P = .02

(Continues)

TABLE 2 (Continued)

Author (y)	Country	n (CHD/ non-CHD)	Study design	Age (y)	CHD DIAGNOSIS CATEGORISED	Assessment	Outcome	Results
Ray et al, 2011 ²⁵	United States	84 (without non-CHD)	CSS-CG	12.0 ± 1.39	Mild CHD ^d (n = 13); Moderate CHD (n = 18); Surgically treated CHD (n = 34); Complex or Severe CHD (n = 19)	Youth Risk Behavior Survey (YRBS); Self-efficacy Instrument (SES).	YBRBS: Risky behaviours and lifestyle habits. SES: Level of self- efficacy specific to PA as perceived by the child. Scale between 8 and 40	Self-efficacy scores: CHD 30.8 vs Healthy Adolescents 29.4. Moderate correlation of Self-efficacy scores with physical activity: $r = .47$; $P < .001$. PA participation was low (38%)
Chen et al, 2012 ²⁶	Taiwan	126 (without non-CHD)	LS	14.6 ± 1.6 (range 12-18)	Class I ^a (n = 48); Class II (n = 20); Others (n = 58)	A modified version of the Child/ Adolescent Activity Log (CAAL)	All basic extents of PA, including frequency, intensity, time, and type	TPA in Summer Vacation vs Fall Semester (min/wk): 806.1 vs 679.2 ($P = .024$); Walk in Summer Vacation vs Fall Semester (min/ wk): 359.4 ± 410.9 vs 265.1 ± 370.9 ($P < .01$); Follow the recommendations of the cardiologist in PA: Summer Vacation 43% vs Fall Semester 50% ($P = ns$)
Moschovi et al, 2019 ²⁷	Greece	154 (76/78)	CSS-CG	12.59 ± 1.28	Class I ^a (n = 22); Class II (n = 48); Class III (n = 6)	Self-reporting instrument (PDPAR); Self-efficacy Instrument (SES).	Intensity, frequency and duration of PA (METs). Self-efficacy. Correlation SES-AF	SES CHD vs CG: 29.1 ± 7.28 vs 29.04 ± 6.6 ($P = .076$); PA (MET) CHD vs CG: 2.37 ± 0.84 vs 3.1 ± 0.95 ($P = .002$); 30min blocks (>3 MET) CHD vs CG: 3.65 ± 2.45 vs 4.85 ± 2.25 ($P = .002$); 30min blocks (>6 MET) CHD vs CG: 1.8 ± 1.82 vs 3.18 ± 2.06 ($P = .001$); Correlation SES to PA: CHD $r = .515$ ($P = .001$); CG $r = .347$ ($P = .002$).

Abbreviations: CG, control group; CHD, congenital heart disease; CSS, cross-sectional study; LS, longitudinal study; NW, normal weight; OB, obesity; OW, overweight; PA, physical activity; PNAD: published normative adolescent data; ST, sedentary time; TPA, total physical activity; VPA, vigorous physical activity.

^aClassification categorised by complexity according to the 2018 AHA/ACC ACHD Guidelines.⁴⁶

^bSeverity according to the criteria of Freed et al.⁴⁷

^cClassification of heart disease according to guidelines published by Gutgesell et al.⁴⁸

^dDiagnostic categories by Uzark et al.⁴⁹

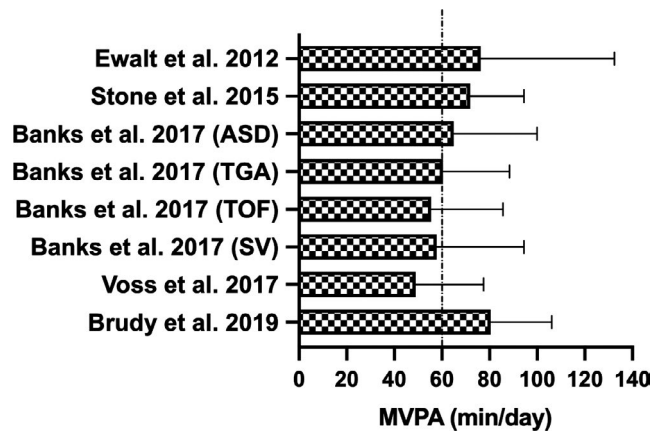


FIGURE 2 Results of moderate-to-vigorous physical activity (MVPA). ASD, atrial septal defect; MVPA, moderate-to-vigorous physical activity; SV, single ventricle; TGA, transposition of the great arteries; TOF, Tetralogy of Fallot. The dotted line indicates the optimal MVPA/day time recommended by WHO

self-reported by the CG was greater than the PA reported by CHD group in both summer and winter.

Pasquali et al²⁴ studied the impact of obesity on the PA of patients with CHD. Through self-reported questionnaires, they collected data regarding the PA restriction and the level of PA performed. They divided the study participants into three groups (normal weight [NW], overweight [OW] and obesity [OB]) and highlighted the non-significant difference between the NW and OW + OB groups in total time of physical activity (TPA), vigorous physical activity (VPA) and inactivity (ST). The total number of hours of ST per week was significantly higher for all the groups in comparison with the total TPA ($P < .001$). The limitation of aerobic activities by the family was significantly higher in the three groups in contrast to the recommendations of the cardiologist.

Ray et al²⁵ evaluated the perception of PA using a self-report survey with eight items and a scale of 8-40 points (higher value, greater self-efficacy). The average score was 30.8 ± 5.7 , and in 5 of 8 questions, more than 75% of the respondents reported high self-efficacy. There was no significant difference according to CHD. That study also evaluated the amount of PA; 38% of the respondents reported 60 minutes of PA, seven days per week; no difference was found between the study group and the CG.

Chen et al²⁶ compared the amount and intensity of exercise in children with mild CHD during summer vacation and during the fall academic semester. Walking (light exercise) was the most frequent activity in both periods. Higher TPA time values were obtained in the summer in comparison with autumn ($P = .024$). The girls that were surveyed performed significantly less moderate PA (MPA) ($P = .019$), VPA ($P < .001$) and TPA ($P = .015$) than their male counterparts. However, in summer, only 43% followed the cardiologist's recommendations regarding the intensity of PA, while 50% did so in the fall.

Moschovi et al²⁷ studied the levels of PA and self-efficacy of Greek children with CHD based on questionnaires and compared them with a CG. It was shown that the children with CHD

participated in lower intensity PA based on average MET. The PA > 6 MET, based on 30-minute blocks, was higher in the CG than the CHD group. No gender differences were observed. PA correlated positively with self-efficacy in both groups. This explained 26.6% of the variability in PA in children with CHD, demonstrating that CHD had a moderate association with PA.

Kao et al²⁸ reported that healthy males perform more high-intensity PA than their peers with CHD during school days ($P = .04$). This same outcome was not found on weekends. Moreover, girls without CHD spent more time in less intense activities than their peers with CHD during the weekend ($P = .01$). There were no differences in PA during the week.

Voss et al²⁹ found that the median self-reported PA in children with CHD (scale 1-5) was somewhat lower than those reported in healthy controls. The MVPA measured objectively was higher in the children that played soccer (55 vs 39 minutes/d, $P = .01$) and in those that swam (57 vs 42 minutes/d, $P = .01$), in comparison with those that did not engage in either activity. The MVPA was significantly higher in the children that participated in competitive sports outside of school (54 vs 39 minutes/d, $P < .01$), with no difference for competitive sports played at school.

3.2 | Direct PA measurement

Among the articles that applied direct PA measurement, one used indirect calorimetry to measure resting energy expenditure (REE) and total energy expenditure (TEE),³⁵ and the other seven used accelerometers²⁸⁻³⁴ with a variable number of measurement days (Figure 2, Table 3). Of the latter, one study used the device for three consecutive days and reported measures of REE,²⁸ while the remaining six evaluated the time of MVPA per day for seven days²⁹⁻³⁴; and two of those studies reported the steps performed per day.^{29,30}

Ewalt et al³⁰ showed that, on average, the total sample exhibited sedentary behaviour 52% of the time, which was significantly less during the weekend, with no difference between the groups. Over the weekend, the MPA was higher and the MVPA was longer for the group with CHD than the CG.

In the article by Stone et al,³¹ children with CHD had less TPA and MVPA and higher ST than the CG ($p = ns$). Only 30% of the children with CHD managed to exceed 60 minutes/d of MVPA.

Banks et al³² showed that MVPA did not differ significantly between groups of children with different types of CHD: atrial septal defect (ASD), transposition of greats arteries (TGA), TOF or single ventricle (SV) ($P = .68$). Moreover, the average PA of the children was similarly distributed by age and gender; the data were obtained using the Canada Health Measures Survey.

Voss et al²⁹ found that the average MVPA and the number of steps per day were below the international recommendations in children with CHD. They showed that only 8% of the children complied with the recommendation of 60 minutes/d of MVPA; there was no difference based on the type of CHD. However, the boys were significantly more active than the girls.

TABLE 3 Summary table of primary articles that incorporate direct measures of physical activity

Author (y)	Country	n (CHD/ non-CG)	Study design	Age (y)	CHD diagnosis categorised ^a	Assessment	Outcome	Results
Leitch et al, 2000 ³⁵	United States	17 (7/10)	CSS-CG	5.7 ± 0.5	Class II (n = 4) Class III (n = 3)	Indirect calorimetry and the doubly labelled water method	REE and TEE	REE (kcal/kg/d) CHD vs CG: 56.9 ± 7.7 vs 58.4 ± 7.7 (P = ns); TEE (kcal/kg/d) CHD vs CG: 69.6 ± 14.4 vs 75.0 ± 8.9 (P = ns); Difference between TEE and REE (kcal/kg/d): CHD: 12.0 ± 11.7; CG: 16.6 ± 10.1, P = ns
Arvidsson et al, 2009 ³³	Sweden	163 (57/106)	CSS-CG	9-11 or 14-16	Class I (n = 15) Class II (n = 27) Class III (n = 15)	ActigReg [®] Activity Monitor (PreMed AS, Ytre Enebakk, Norway)	TPA, MVPA	Only four children (2.4% from CHD and CG) achieve 60 min MVPA (1 CHD vs 3 CG). In general, patients and controls perform a similar MVPA time (mean MVPA/d: 26 min)
Kao et al, 2009 ²⁸	Taiwan	68 (34/34)	CSS-CG	10.5 ± 1.0 (range 9-12)	Class I (n = 23) Class II (n = 11)	Triaxial Research Tracker (RT3) (Stayhealthy Inc.)	TEE and PAL	TEE boys wk (kcal/d) CHD vs CG: 1657.5 ± 285.2 vs 1968.3 ± 278.5 (P = .003); TEE Boys wknd (kcal/ day) CHD vs CG: 1544.5 ± 232.2 vs 1823.8 ± 260.8 (P = .002); TEE Girls wk and wknd there are no differences between groups
Ewalt et al, 2012 ³⁰	United States	42 (21/21)	CSS-CG	10.7 ± 3.2 (range 6.6-17.1)	Class I (n = 2) Class II (n = 11) Class III (n = 8)	Accelerometer Actigraph 7164 (Actigraph, LLC)	PA, intensity (ST, MPA, VPA, MVPA)	52% of the time in sedentary lifestyle. No difference between groups; Sedentarism wk vs wknd (min/d): CHD 423.9 ± 120.8 vs 348.0 ± 100.2 (P < .05); CG 418.0 ± 94.2 vs 366.9 ± 106.7 (P < .05), no difference between groups. MVPA total (min/d) CHD vs CG: 76.3 ± 56.1 vs 56.1 ± 28.1 (P = .085); MVPA wk (min/d) CHD vs CG: 73.0 ± 49.8 vs 59.8 ± 29.9 (P = .251); MVPA wknd (min/ day) CHD vs CG: 73.9 ± 59.2 vs 45.7 ± 30.8 (P = .048)
Stone et al, 2015 ³¹	Canada	20 (10/10)	CSS-CG	4.0 ± 1.0 (range 3-5)	Class II (n = 10)	Accelerometer Actigraph GT1M (Actigraph, LLC)	Total time of ST, TPA, LPA, MPA, VPA and MVPA	CHD vs CG (min/d): ST: 446.8 ± 48.7 vs 429.4 ± 37.8; TPA: 219 ± 39.9 vs 224.1 ± 44.0; LPA: 147.5 ± 22.3 vs 143.8 ± 21.7; MPA: 44.0 ± 11.8 vs 48.1 ± 12.7; VPA: 27.9 ± 11.7 vs 32.2 ± 13.2; MVPA: 71.9 ± 22.6 vs 80.3 ± 24.5
Banks et al, 2017 ³²	Canada	137 (without non-CHD)	CSS	7.7 ± 2.0 (range 4-12)	Class I (n = 31) Class II (n = 37) Class III (n = 69)	Accelerometer (Respironics Actical 2.1)	MVPA min/wk	MVPA does not vary significantly between CHD groups (P = .68); ASD: 454 ± 246 min/wk; TGA: 423 ± 196 min/wk; TOF: 389 ± 211 min/wk; SV: 405 ± 256 min/wk
Voss et al, 2017 ²⁹	Canada	90 (without non-CHD)	CSS	13.6 ± 2.7	Class I (n = 22) Class II (n = 28) Class III (n = 31) Heart Transplant Questionnaire (PAQ) (n = 9)	Accelerometer Actigraph GT3X+, GT9X (Actigraph LLC), PA Questionnaire (PAQ)	Sedentary time (%/d), intensity of PA (min), steps. PAQ: measures participation in different types of PA. Scale between 1-5	Mean MVPA: 49.0 ± 28.5 min/d; Median MVPA: 42.6 (28.9-56.9) min/d; Median VPA: 12.4 (6.1-20.7) min/d; Median TPA: 306.0 (243.1-417.9) counts (×10 ³)/d; Steps 7,494 (6418-9892) count/d; Sedentary time 70.0 (61.2- 75.9) %/d. There was no difference in MVPA between those with and without activity restriction (P > .05). Median PAQ: 2.6 (CI 2.1-3.1)

(Continues)

TABLE 3 (Continued)

Author (y)	Country	n (CHD/ non-CG)	Study design	Age (y)	CHD diagnosis categorised ^a	Assessment	Outcome	Results
Brudy et al, 2020 ³⁴	Germany	258 (162/96)	CSS-CG	11.4 ± 3.4 (range 5.8-17.6)	Class I (n = 29) Class II (n = 58) Class III (n = 74) Miscellaneous (n = 1)	Accelerometer Garmin Vivofit Jr (Garmin Ltd)	MVPA and Steps per day	Whole Week MVPA/day adjusted (CHD vs CG): 80.5 ± 25.6 vs 81.5 ± 25.3 (P = .767); MVPA wk (min/d) CHD vs CG: 83.6 ± 27.7 vs 83.5 ± 28.9 (P = .987); MVPA wknd (min/day) CHD vs CG: 72.8 ± 33.8 vs 76.8 ± 35.2 (P = .405); Whole Week Steps/day adjusted (CHD vs CG): 10 206 ± 3178 vs 11 142 ± 3136 (P = .04); Steps/day wk (CHD vs CG): 10 717 ± 3525 vs 11 372 ± 3664 (P = .191); Steps/day wknd (CHD vs CG): 8917 ± 4072 vs 10 604 ± 4242 (P = .004)

Abbreviations: ASD, atrial septal defect; CG, control group; CHD, congenital heart disease group; CSS, cross-sectional study; LPA, light physical activity; MPA, moderate physical activity; MVPA, moderate-to-vigorous physical activity; PA, physical activity; PAL, physical activity level; REE, resting energy expenditure; ST, sedentary time; SV, single ventricle; TEE, total energy expenditure; TGA, transposition of the great arteries; TOF, tetralogy of Fallot; TPA, total physical activity; VPA, vigorous physical activity; wk, week; wknd, weekend.

^aClassification categorised by complexity according to the 2018 AHA/ACC ACHD guidelines.⁴⁶

Arvidsson et al³³ reported that only 2.4% of the total sample managed to exceed 60 minutes/d of MVPA, and there were no differences between the groups, which averaged 26 minutes/d. However, in the group of 9- to 11-year-olds, a moderate-to-heavy total level of PA prevailed, in contrast to the group of 14-16 years old in which the mild-to-moderate level predominated.

Brudy et al³⁴ observed that 75.9% of the CHD group and 84.3% of the CG performed more than 60 minutes/d of MVPA (P = .767). Although the CHD group accumulated more than 10 000 steps daily, it performed -936 ± 454 steps compared to the CG (P = .04).

In contrast to previous articles, Leitch et al³⁵ measured the impact of heart disease on PA during one week, based on the difference between resting REE and TEE through doubly marked water. On average, children with CHD had a lower REE and a lower TEE (P = ns) than the CG. The difference in the energy spent on PA was not significant between the two groups.

Kao et al²⁸ evaluated PA in children with CHD and compared them with a CG. They found that both the TEE and the level of PA in boys were lower in the CHD group. However, no differences in the TEE and PA level were found for the girls. The MVPA time was similar in both groups (P = .43).

4 | DISCUSSION

The main findings of this systematic review are that there are a variety of ways to measure PA in children with CHD. In the articles that objectively evaluated PA, MVPA was the outcome that was most often measured, showing that the subjects with data available for analysis and affected by a CHD do not reach the threshold of 60 minutes/d of MVPA recommended by WHO.

Although the study by Brudy et al³⁴ reported high values of MVPA for children with CHD, and it had the largest sample size of the set of articles, its results should be viewed with caution in view of its methodological quality assessment, ranking within the last three places of compliance.

Recommendation guidelines state that, in order to obtain a more reliable magnitude of PA, it must be measured objectively for at least seven days, due to the variability between weekdays and weekends, especially considering children attending different levels of children in different grades in school.^{36,37} However, the selected articles that used an accelerometer and reported MVPA²⁹⁻³⁴ showed differences in the number of days registered under this method. Although the six studies declared the intention to measure PA for seven consecutive days, all of them considered it to be acceptable to only measure PA for three days a week, and four of the studies^{30-32,34} added a weekend day. Of the latter, however, only two reported their differences.^{30,34}

Another way to recommend and evaluate PA is by measuring the number of steps/day. To quantify the steps, in addition to accelerometers, pedometers have been employed due to their good reliability, low cost and ease of use.³⁸ While WHO recommends walking 10 000 steps/day to maintain health,¹⁰ in 2004,

Tudor-Locke et al³⁹ published pedometer data in healthy subjects ranging in age between 6 and 12; they concluded that boys with <15 000 steps/d and girls with <12 000 steps/d were more likely to be classified as overweight or obese according to their body mass index. However, in 2017, Oliveira et al⁴⁰ changed the paradigm by focusing the analysis on the achievement of the recommendations of 60 minutes/d of MVPA and performing the measurement with an accelerometer. Thus, the cut-off points were determined to be 9703 steps/day for boys and 9445 steps/day for girls. In our review, only Voss et al and Brudy et al^{29,34} reported this variable, showing that walking an average of 8900 steps is well below the number of steps recommended by the WHO international guidelines (11% less). Recent publications have also given importance to the cadence (steps/min) for paediatric ages, indicating that the guideline for moderate PA is 128.4 steps/min and the guideline for vigorous PA is 157.7 steps/min.⁴¹ However, not all devices offer this information, so their use would be limited.

Although there is evidence that between childhood and adolescence PA decreases in healthy subjects,⁴² only three articles^{28,30,33} characterise the sample by age range, of these, only one³³ reports its results separately, coinciding with the previously available evidence. However, to obtain more robust conclusions, it is necessary to compare these results between subjects from the same population or with a CG.

One of the factors that determine the level of PA has to do with the social environment and the influence that the family has on children.⁴³ In the case of children with CHD, in addition to the limitation in physical capacity due to the heart defect,¹³ the social restriction is even greater and probably maximised by the parents.⁴⁴ Several authors showed that the restriction for the development of PA has to do with aspects surrounding cardiac dysfunction, such as the restrictive attitude of the family limiting the ability of follow the recommendations of the cardiologist,^{22,28} and the self-efficacy that the child perceives of his/her own ability to perform PA.²⁷

The severity of CHD is another aspect to consider within the determinants of PA. Although some articles did not address that association^{23,29,31,32} and others affirm it,^{21,30,34} there is a consensus that people with univentricular pathologies have a higher risk of presenting lower PA values than their peers with less complexity. This needs to be confirmed more precisely in future studies.

Most of the articles that measured PA through questionnaires used self-report tools. Although they are affordable and easy to apply, they are an indirect measurement that uses subjective data, requires a complex process of validation and only evaluates the most common activities, omitting the infrequent activities that might involve a large energy expenditure.⁴⁵ Furthermore, a high heterogeneity was observed in the tools used for this purpose, both in quality (five validated,²³⁻²⁷ three not validated²⁰⁻²²), as in the way of reporting the results were reported, favouring global measures of PA instead of MVPA, which is a unit of measurement widely recommended by the WHO.

Although the indirect measurement instruments were intended to measure PA, all the articles did so using a different questionnaire.

This included directed sports participation surveys,^{20,21} self-reports of participation in pre-defined activities over a period of time^{22,24-26} or the day before²⁷ and instruments for the national assessment of PA.²³ This made it impossible to compare the PA findings between studies, so our conclusions focused on the results obtained from objective evaluations.

Of the 16 articles that were ultimately selected, only six measured the MVPA. This may be due to multiple factors such as the cost of the devices that measure it, which are generally more expensive since they incorporate the use of accelerometers and software for their interpretation.⁴⁵ However, most of these devices have been used in the last decade. Still, it important not only to consider the instruments that objectively measure the MVPA but also to consider the use of questionnaires. In this way, accelerometers or questionnaires can be used depending on the available resources and the objective of the measure.

Within the limitations of the study, we mentioned the great variety of diseases described in the primary articles that, due to their anatomical and functional variability, prevent the ability to obtain less biased conclusions about the impact of the cardiac structural malformation on PA. Moreover, the variability of age ranges makes it difficult to reach conclusions about the impact of age with a low level of bias.

Finally, studies that consider populations with narrower age ranges that include more homogenous diseases and that consider adequate sample sizes to obtain more robust conclusions on the evaluation of PA are an essential dimension in the follow-up and prognosis of this paediatric population. In this sense, we recommend the use of reliable and sensitive tools, such as accelerometers, to evaluate PA.

In conclusion, there are a variety of ways to measure PA in children with CHD. In the articles that objectively evaluated PA, the most measured outcome was MVPA, showing that the MVPA time for 46% of children with CHD is less than what is recommended by WHO. Additionally, we found that children with CHD took fewer steps/day than is recommended. In this context, the family restriction to PA plays a fundamental role in the children's behaviour. A sedentary lifestyle is common in this population, and it could harm the children's health and quality of life.

CONFLICT OF INTEREST

The authors declare no conflicts of interest.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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