Objective Physical Activity Assessment in Clinical Congenital Heart Disease Research: A Systematic Review on Study Quality, Methodology, and Outcomes

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Keywords

Accelerometers · Congenital heart disease · Physical activity · Prevention exercise

Abstract

Background: The shift toward a preventative approach in medical aftercare of congenital heart disease (CHD) patients has led to encouragement of regular physical activity (PA) in this patient population. Objective measures are crucial in accurately displaying PA levels and have increasingly found their way into clinical research. This review aims to give an overview about quality, methodology, and outcomes of current scientific work on accelerometers objectively assessing PA in patients with CHD. **Methods:** Systematically researched literature in all relevant databases (PubMed, Cochrane, and Scopus) over the past decade (2009–2019) with history of CHD and accelerometer-based PA assessment was evaluated by 2 independent reviewers according to the Study Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies of the National Heart, Lung, and Blood Institute. **Results:** Eight articles with 664 pediatric patients with CHD aged 3–18 years (range 10–162 patients), 5 studies with 574 adults with CHD aged 18–63 years (range 28–330 pa-

tients), and 3 studies with 177 pediatric patients and adults with CHD aged 8–52 years were included. Two studies were rated “good”; 9, “fair”; and 5, “poor.” Methodologies and devices differed substantially across all studies. **Conclusions:** Overall study quality was fair at best, and due to difficult methodological comparability of the studies, no clear answer on how active patients with CHD really are can currently be given. Larger studies carefully considering collection and processing criteria, and correct reporting standards exploring PA in patients with CHD from different angles are needed.

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Introduction

Regular physical activity (PA) has long been known as one of the most effective preventative tools for a vast number of diseases and clinical conditions, especially those of cardiovascular origin [1, 2]. Patients with congenital heart disease (CHD) have inhibition to different extends within their cardiovascular system since birth, and with increased survival, medical aftercare has shifted toward prevention of secondary diseases in this patient

Fig. 1. Search and selection process for systematic review according to Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).

population [3]. Part of this preventative approach is the encouragement to regular PA and sports participation [4] as it is now considered beneficial without additional risk for almost all patients with CHD. The encouragement toward a generally physically active lifestyle becomes imperative in this context [3]. Technological advances offer exciting research opportunities [5] and since subjectively estimated PA fails to draw accurate pictures [6], objective measures to assess PA have also found their way into clinical CHD research. After more than a decade of research assessing PA with accelerometers in CHD patients, it is time to evaluate where the field is at and where it needs to go from there. Therefore, this review aimed to give an overview about quality, methodology, and outcomes of current scientific work on accelerometers objectively assessing PA in CHD patients.

Material and Methods

Search Strategy

A systematic literature research referring to January 2009 until December 2019 was conducted in the electronic databases PubMed, Cochrane, and Scopus. Two independent reviewers identified rel-

evant original articles and randomized controlled trials (RCT) in English language. A standard protocol with search terms was generated according to the population, intervention, comparison, outcome, context (PICO-C) [7] method and connected as follows:

1. Congenital heart disease OR congenital heart diseases OR congenital heart defect OR congenital heart defects.
2. Physical activity assessment OR physical activity measurement OR physical activity OR daily activity OR accelerometer OR accelerometry OR wearable OR activity tracker OR activity monitor OR fitness tracker.

Medical Subject Headings terms and similar filters (clinical trial, randomized controlled trial, published in the last 10 years, humans, English language) were used and appropriately adapted if necessary. According to the latest PRISMA statement [8], supplementary reference lists were analyzed to detect further eligible articles.

Data Collection

Both reviewers screened all relevant articles for titles and abstracts which had to fulfill the basic inclusion criteria: history of CHD and accelerometer-based PA assessment. PA had to be classified in some sort of PA intensity and not merely step count alone as moderate-to-vigorous physical activity (MVPA) is shown to be beneficial for cardiovascular health [9].

At least one of the two reviewers had to consider a reference eligible. A third reviewer was consulted to resolve disagreement before and during full-text analysis.

Table 1. Quality assessment according to the NHLBI Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies

Study	Type	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Q12	Q13	Q14	Quality rating
<i>Pediatric CHD (n = 8)</i>																
Kao et al. [18]	CSS	✓	✓	✓	-	-	✓	-	-	✓	NA	✓	-	✓	-	Poor
Ewalt et al. [19]	CSS	✓	✓	✓	✓	✓	-	-	NA	✓	-	✓	-	✓	-	Fair
Longmuir et al. [20]	RCT	✓	✓	✓	✓	✓	✓	✓	-	✓	✓	✓	✓	✓	✓	Good
Stone et al. [21]	CSS	✓	✓	✓	-	-	-	-	NA	-	-	✓	-	✓	-	Poor
Klausen et al. [22]	RCT	✓	✓	✓	✓	✓	✓		-	✓	✓	✓	✓	✓	✓	Good
Banks et al. [23]	CSS	✓	✓	-	-	-	✓	✓	✓	✓	-	✓	-	✓	-	Poor
Voss et al. [24]	CSS	✓	-	CD	✓	-	✓	-	NA	✓	-	✓	-	✓	✓	Poor
Brudy et al. [25]	CSS	✓	✓	-	✓	-	✓	✓	✓	✓	-	✓	-	CD	✓	Fair
<i>ACHD (n = 5)</i>																
Dua et al. [26]	CCT	✓	✓	-	✓	-	✓	✓	✓	✓	✓	✓	-	✓	-	Fair
Müller et al. [27]	CSS	✓	✓	-	✓	-	-	-	NA	✓	-	✓	-	✓	✓	Fair
Sandberg et al. [28]	CSS	✓	✓	✓	✓	-	✓	-	NA	✓	-	✓	-	✓	✓	Fair
Opatowsky et al. [29]	RCT	✓	✓	✓	✓	-	✓	✓	-	✓	✓	✓	-	✓	✓	Fair
Larsson et al. [30]	CSS	✓	✓	✓	✓	✓	✓	-	✓	✓	-	✓	-	✓	-	Fair
<i>Pediatric and ACHD (n = 3)</i>																
Müller et al. [31]	CSS	✓	✓	✓	✓	✓	-	-	-	✓	-	✓	-	✓	-	Fair
Duppen et al. [32]	RCT	✓	✓	-	-	✓	✓	✓	-	✓	✓	✓	-	✓	-	Fair
Hedlund et al. [33]	CCS	✓	-	✓	-	-	-	-	NA	✓	-	✓	-	✓	-	Poor

CD, cannot determine; NA, not applicable; ✓ denotes “yes”; - denotes “no”; RCT, randomized controlled trial; CCT, controlled clinical trial; CSS, cross-sectional study; CHD, congenital heart disease; ACHD, adults with congenital heart disease; NHLBI, National Heart, Lung, and Blood Institute.

Both reviewers rated the included literature according to the Study Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies of the National Heart, Lung, and Blood Institute [10] – a 14-item list assessing potential risk for bias – and consequently categorized them as good, fair, or poor.

Results

Description of Selected Studies

Figure 1 displays the search and selection process. Reasons for exclusion after full-text analysis were: One study protocol [11], two validation studies [12, 13], four studies without reporting PA intensity [14–17]. Overall, 16 articles with a total of 1,415 CHD patients met all inclusion criteria.

Study Characteristics

Eight studies [18–25] evaluated 664 pediatric patients with CHD aged 3–18 years (range 10–162 patients), 5 stud-

ies [26–30] 574 ACHD aged 18–63 years (range 28–330 patients). Three studies [31–33] with 177 patients (range 30–99 patients) examined both pediatric patients and adults with CHD aged 8–52 years but did not report results separately for these age ranges. Detailed information on study types and quality rating can be found in Table 1.

The majority of cross-sectional studies (CSS) compared their CHD population to a healthy reference cohort (RC) [18, 19, 21, 25, 28, 30, 33]; the remaining [23, 24, 27, 31] compared PA within their CHD sample. All but 4 studies [20, 22, 23, 29] compared their patients' PA to a healthy RC, against varying institutional PA recommendations, or against both. All CSS evaluated total daily (and where applicable weekly) PA, MVPA, or separate PA intensities including sedentary time. Subjective PA was also compared in seven CSS [18, 21, 25, 30, 33]; three CSS [23, 27, 31] and one RCT [32] examined QoL; and three CSS exercise capacity parameters [23, 27, 31] in relation to PA.

Intervention studies lasted 12-months in pediatric patients with CHD [20, 22], 12 weeks in adolescents and

Table 2. Study characteristics and outcomes

Study	CHD, <i>n</i> (female)	Healthy CG (<i>n</i>)	CHD diagnosis (<i>n</i>)	Age ± SD (range)	Outcome measures intensity classification	Guideline	Results
<i>Pediatric CHD (n = 8)</i>							
Kao et al. (2009) [1]	34 (17)	Age & gender matched (34)	VSD (16), ASD (7), ToF (11)	10.5 (9–12)	Objective and subjective TEE, average total daily EE, EE in MVPA PAL, ² subjective MVPA Schofield (1985) ³	PAL > 1.63 Megajoule/ day (0.025 × age + 1.40) Hoos et al. (2003) ⁴	No difference in MVPA between pediatric CHD (188.6 ± 181.9) and RC (223.4 ± 201.6, <i>p</i> = 0.60), also not on weekends (180.1 ± 235.8 vs. 154.0 ± 158.5, <i>p</i> = 0.71). TEE and PA lower for boys with than without CHD. TEE did not differ between girls with and without CHD. Girls with higher PA than boys
Ewalt et al. (2012) [5]	21 (16)	Sex & grade-in- school matched (21)	CoA (5), hypoplastic left heart (4), VSD (3), ASD (1), D-TGA (1), PS (1), Bicuspid aortic valve & stenosis (1), mitral valve prolapse (2), DILF (1), ToF (1), Tricuspid atresia (1)	10.7 ± 3.2 (6.6–17.1)	Time in total PA: total counts/30 s/day, SED: ≤ 50 counts per 30 s period. Moderate PA: 4.0–6.9 MET, vigorous PA: ≥ 7 MET. Average daily PA. Freedson et al. (2005) ⁶	≥ 60 min of moderate PA and/or vigorous PA/day American Alliance for Health, Physical Education, Recreation and Dance ⁷	No difference in PA min (287.2 ± 110.1 vs. 262.0 ± 57.5, <i>p</i> = 0.23), MPA (57.5 ± 37.9 vs. 50.7 ± 23.9, <i>p</i> = 0.26), or VPA (13.1 ± 12.4 vs. 9.9 ± 5.9, <i>p</i> = 0.28) in CHD versus CG. 20% of CHD reached ≥ 60 min of moderate PA and/or vigorous PA/day. Majority of time (52%), 6.7 of 13 monitored hours) spent in SED. > 1/5 of all reach ≥ 60 MVPA min/day
Longmuir et al. (2013) [8]	61 (25)	–	All after the Fontan procedure	9.1 [IQR: 7.7; 10.5]	Daily MPVA as sum of all counts exceeding 400 per 15-s epoch min/week Gross motor development; health-related fitness, hand grip strength, hamstring flexibility, CPET, activity attitudes Puyau et al. (2004) ⁹	–	MVPA increased to 36 ± 31 min/week (<i>p</i> = 0.04) above baseline and was sig. Greater (<i>p</i> = 0.03) than predicted age decrease at 2-year FU in IG; no association between weekly MVPA min and diagnosis (<i>p</i> = 0.29), left-or-right ventricle physiology (<i>p</i> = 0.55), years since Fontan operation (<i>p</i> = 0.76), or use of ACE inhibitors (<i>p</i> = 0.31); no difference in MVPA in rural versus urban patients (<i>p</i> > 0.05)
Stone et al. (2015) [10]	10 (4)	Age-, sex-, data- acquisition- season- matched (10)	CoA (6), ToF (4)	4.0 ± 1.0 (3–5)	SED: < 8 counts/3 s, light PA: 8–83 counts/3 s, moderate PA: ≥ 84 and < 168 counts/3 s, MVPA: ≥ 84 counts/3 s, vigorous PA: ≥ 168 counts/3 s Parent-estimated PA habits of child Pate et al. (2006) ¹¹	≥ 180 min of any PA type PA/day Canadian physical activity guidelines for the early years ¹²	No difference in total (219 ± 39.9 vs. 224.1 ± 44.0, <i>p</i> = 0.80), light (147.5 ± 22.3 vs. 143.8 ± 21.7, <i>p</i> = 0.71), moderate (44.0 ± 11.8 vs. 48.1 ± 12.7, <i>p</i> = 0.46), MVPA (71.9 ± 22.6 vs. 80.3 ± 24.5, <i>p</i> = 0.43), or vigorous (27.9 ± 11.7 vs. 32.2 ± 13.1, <i>p</i> = 0.45) PA min/ day between pediatric CHD and CG. Only 40% of CHD and CG reached ≤ 180 min PA/ day. 90% of parents reported their children as active as healthy, and 80% of parents 1– 3 h daily screen time
Klausen et al. (2016) [13]	158 (66)	–	CoA (52), TGA (35), Fallot (21), DORV (7), Truncus arteriosus (4), AVSD (9), TCPC (6), other (24)	14.6 ± 1.3 (13–16)	Total MVPA min/day: > 2,000 accelerometer cut point counts/min CPET –	–	No difference in MVPA min/day between IG (40.3 ± 21.8) and CG (41.3 ± 22.9). 32% of IG and 26% of CG reached ≥ 60 MVPA min/ day at baseline Adding tailored eHealth did not affect outcomes in adolescents with CHD

Table 2 (continued)

Study	CHD, <i>n</i> (female)	Healthy CG (<i>n</i>)	CHD diagnosis (<i>n</i>)	Age ± SD (range)	Outcome measures intensity classification	Guideline	Results
Banks et al. (2017) [14]	137 (59)	–	ASD (31), TGA after Switch (34), ToF (37), Fontan (35)	8.5±2.1 (4–12)	Total daily and weekly MVPA >1,600 counts/min CPET, gross motor skill, QoL Puyau et al. (2004) ⁹	–	No difference in MVPA between CHD groups ($p = 0.68$). Higher MVPA was associated with higher overall self-efficacy ($p = 0.02$) and trend toward higher %-predicted VO_2 ($p = 0.052$) but not RER, CHD type or gross motor skill percentile ($p > 0.05$ for all)
Voss et al. (2017) [15]	81 (34)	–	<i>Mild</i> (26): 6 Bicuspid aortic valve; 5 VSD, 4 AS, 4 ASD, 3 mixed aortic valve disease, 2 PS, 1 mitral valve disease, 1 PDA <i>Moderate</i> (26): 11 ToF, 7 CoA, 2 AS, 1 total anomalous pulmonary venous return, 1 ostium primum ASD, 1 sinus venous ASD, 1 EBS, 1 Shone syndrome, 1 Pentalogy of Cantrell <i>Severe</i> (29): 17 Fontan, 8 TGA, 2 DORV, 2 valved conduit <i>Cardiac Transplants</i> (9)	13.6±2.7 (8–19)	Total PA: sum of vertical acceleration counts, MVPA: ≥2,296 CPM, vigorous PA: ≥4,012 CPM, % of SED time relative to valid accelerometer wear time, subjective PA Evenson et al. (2008) ¹⁶	60 min of MVPA on ≥6 days/week Canadian health measures Survey ¹⁷	No difference in various PA intensities between CHD severity groups. Boys were sig. More active than girls (49.2 [33.1–67.2] vs. 39.2 [25.1–46.9], $p = 0.006$). Age (–3.2 [–5.3, –1.1] $p < 0.01$) and sex (–14.7 [–25.9, –3.5], $p < 0.05$) related declines in MVPA min/day Median daily MVPA was 43 (IQR: 28.9–56.9) min/day. 8% (12% boys, 4% girls) reached ≤60 MVPA min on ≤6 days/week
Brudy et al. (2019) [18]	162 (60)	Yes (96)	29 simple, 58 moderate 74 complex, 1 not defined Left heart obstruction (28), right heart obstruction (53), Isolated shunts (20), TGA after Switch (19) TCPC (25), Miscellaneous (17)	11.8±3.2 (5.8–17.6)	Daily steps and MVPA, subjective PA	≥60 MVPA min/day WHO ¹⁹	No difference in daily MVPA min (80.5±25.6 vs. 81.5±25.3, $p = 0.767$), only lower step count (10,206±3,178 vs. 11,142±3,136, $p = 0.040$) between pediatric CHD and CG. 76% CHD and 84% CG reached ≥60 MVPA min/day ($p = 0.217$) while age ($p = 0.004$), higher BMI ($p = 0.002$), overweight & obesity ($p = 0.016$) and complex CHD severity ($p = 0.046$) were associated with not reaching it. Subjective estimation fairly correct in about half the patients
ACHD ($n = 5$) Dua et al. (2010) [20]	61 (17)	–	NYHA class I (21) NYHA class II (16) NYHA class III & IV (13)	31.7±10.9 (18–63)	Objective PA as accelerometer counts/hour. MVPA: >2,100 CPM, activity associated EE, subjective PA QoL, treadmill exercise test	5 × 30 min MVPA/week UK national guidelines for PA ²¹	MVPA min/day increased from 21.9±17.1 to 39.1±27.0 ($p < 0.001$), 14% reached 5×30 MVPA min/week at baseline and 56% at FU (Group I: 23.9–80.9%, group II: 12.6–37.6%, group III: 0–38.5%). All groups sig. increased subjective & objective PA, QoL, treadmill test duration, and AAE

Table 2 (continued)

Study	CHD, <i>n</i> (female)	Healthy CG (<i>n</i>)	CHD diagnosis (<i>n</i>)	Age ± SD (range)	Outcome measures intensity classification	Guideline	Results
Müller et al. (2012) [22]	330 (149)	–	Native or palliated Cyanotic (16), Fontan (42), TGA after atrial switch (59), TGA no atrial switch (13), EBS (7), ToF (56), PS (18), CoA (23), AS (37), Isolated Shunts (35), other (14)	30.0±8.4 (18–61)	Daily PA: 7-day mean activity units. Daily moderate: 3–6 MET, >984 CPM, vigorous: >6 MET, >2,340 CPM, and MVPA: sum of activity >3 MET QoL, CPET Rowlands et al. (2004) ²³	≥30 MVPA min/day Center for disease Control & prevention ²⁴	59.2±39.7 of ≥ moderate PA min/day, 76% reached ≥30 MVPA min/day. Moderate relationship between daily activity and exercise capacity ($r = 0.437$, $p < 0.001$), poor between QoL and PA ($r = 0.030$ – 0.258 , $p > 0.05$ for all). Reduced exercise capacity in most patients (73.7±19.5% of predicted)
Sandberg et al. (2016) [25]	80 (32)	Age & sex matched (42)	Simple (40): 12 CoA, 3 AS, 3 aortic regurgitation, 4 AS, 14 VSD, 1 ASD, 1 PFO, 1 PDA, 1 mitral regurgitation Complex (40): 5 TGA, 2 ccTGA, 8 ToF, 3 PA, 2 DORV, 1 DILV, 7 TCPC, 2 Fontan, 3 EBS, 6 Eisenmenger, 1 miscellaneous	Simple: 30.0 (20.7) Complex: 33.7 (29.4) Median (IQR)	Total accelerometer counts/ day, MVPA min/day No classification	≥150 MVPA min/week – averaged to 21.4 MVPA min/day WHO ¹⁹	No difference in MVPA or in reaching PA recommendations between ACHD and CG or according to NYHA class. 54% of simple, 45% of complex CHD, and 56% of CG reached ≥21.4 MVPA min/day ($p > 0.05$ for all). Simple CHD (107.7 [76.6, 139.1]) had more accelerometer counts/day than complex (72.8 [49.2, 101.0], $p \leq 0.001$), CG (78.3 [58.7, 106.9], $p = 0.002$) Impaired NYHA class predicted PA better than CHD complexity
Opotowsky et al. (2018) [26]	28 (14)	–	ToF (3), TGA (9), Fontan (2), pulmonary atresia (2), truncus arteriosus (1), EBS (1)	41.1±12.1 (29–53)	Daily vector magnitude and counts, SED, light PA, MVPA, MVPA % CPET, QoL, self-reported health status, blood markers No classification	–	No difference in MVPA min/day, step count or estimated MET between groups at baseline and various FU ($p > 0.05$ for all). Cardiac rehabilitation is safe and associated with improvements in aerobic capacity and subjective health status
Larsson et al. (2019) [27]	75 (29)	Age & sex matched (42)	Simple (39), complex (36)	37.5±15.5 (22–43)	Time spent at MVPA (≥3 MET), subjective PA No classification	≥150 MVPA min/week – averaged to 21.4 MVPA min/day WHO ¹⁹	No difference in proportion reaching ≥21.4 MVPA min/day (48.0 vs. 57.1%, $p = 0.34$) between ACHD and CG, or between simple and complex CHD (53.8 vs. 41.7%, $p = 0.29$). ACHD and CG overestimate weekly PA level
<i>Pediatric and ACHD (<i>n</i> = 3)</i>							
Müller et al. (2009) [28]	57 (18) 28 ≤14 & 29 ≥14 years	–	DILV (21), Tricuspid atresia (15), hypoplastic left heart syndrome (11), hypoplastic right heart syndrome (5), DORV with imbalanced ventricles (3), AVSD with imbalanced ventricles (2)	14 [IQR: 11.0; 17.8] (8–52)	Daily activity: mean value of activity units over 3 days. Time in moderate: 3–6 MET, >970 CPM and vigorous PA: >6 MET, >2,333 CPM CPET, QoL Rowlands et al. (2004) ²³	≥60 min, ≥3 MET, ≥5 days/week UK expert consensus group ²⁹	Patients spent 87 (48–112) MPA and 12 (6–24.5) VPA min/day. 72% of CHD patients reached ≥60 min, ≥3 MET, ≥5 days/week. MVPA was associated with age ($r = -0.506$, $p < 0.001$) and VO_{2peak} ($r = 0.432$, $p = 0.001$) daily PA was related to QoL in children <14 years ($r = 0.380$, $p = 0.046$)
Duppen et al. (2015) [30]	99 (66)	–	ToF (47), Fontan (43)	15±3 (10–25)	Counts/min. Converted to MET, time in sedentary, moderate, vigorous, and very vigorous activity CPET Freedson et al. (2005) ⁶	≥60 MVPA min/day WHO ¹⁹	MVPA was 104±65 min/day (13.6±8.6% of measured time) and did not change in IG or CG. 70% reached ≥60 MVPA min/day at baseline and FU. Male patients (69.9±44.8) were more active than female (46.3±27.6; $p < 0.001$) Peak VO_2 and workload increased in IG but not CG. Sig. Increase in TOF but not Fontan patients in VO_{2peak}

Table 2 (continued)

Study	CHD, n (female)	Healthy CG (n)	CHD diagnosis (n)	Age ± SD (range)	Outcome measures intensity classification	Guideline	Results
Hedlund et al. (2016) [31]	30 (14)	Age & sex matched (25)	Fontan (30)	14.2±3.2 (8.9–20.4)	Total vector magnitude. Sedentary: 0–100 CPM, light: 101–2,292 CPM, moderate: 2,293–4,008 CPM, or vigorous: ≥4,009 CPM, subjective physical exercise QoL Evenson et al. (2008) ¹⁶	≥60 MVPA min/day WHO ¹⁹	No difference in MVPA min/day (148±60 vs. 141±54, $p = 0.67$), less time in SED (48.6±4.4 vs. 51.8±5.2%, $p < 0.05$), more light PA (41.1±3.3 vs. 38.4±3.6%, $p < 0.01$), similar MPa (10.3±4.2 vs. 9.8±3.7%, $p = 0.67$) between CHD patients and CG. Age-related decrease in MVPA in patients ($R^2 = 0.18$, $p < 0.05$). 90% of patients reached ≤60 MVPA min/day Patients subjectively reported less time exercising and lower QoL than CG

CHD, congenital heart disease; ACHD, adults with congenital heart disease; NYHA, New York Heart Association; CoA, coarctation of the aorta; TGA, transposition of the great arteries; DORV, double-outlet right ventricle; DILV, double-inlet left ventricle; ASD, atrial septal defect; TCPC, total cavopulmonary connection; ToF, Tetralogy of Fallot; EBS, Ebstein anomaly; AS, aortic stenosis; PS, pulmonary stenosis; PDA, patent ductus arteriosus; PFO, persistent foramen ovale; cc-TGA, congenital-corrected TGA; CTP, cardiac transplant patients; CG, control group; IG, intervention group; SD, standard deviation; SED, sedentary behavior; QoL, quality of life; PA, physical activity; MVPA, moderate-to-vigorous PA; MPA, moderate PA; VPA, vigorous PA; TEE, total energy expenditure; CPET, cardiopulmonary exercise test.¹ Kao CC, Chang PC, Chiu CW, Wu LP, Tsai JC. Physical activity levels of school-age children with congenital heart disease in Taiwan. *Appl Nurs Res*. 2009;22(3):191–7.

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young adults with CHD [32], and 10 or 12 weeks in ACHD [26, 29]. Alongside increasing PA these studies also focused on other parameters beyond of the scope of this review (exercise capacity, QoL, and gross motor skill). Table 2 provides further information on cohort description, outcome measures, and PA classifications.

Accelerometers Used

Detailed information on technicalities and characteristics of the devices used in all included studies can be found in Table 3. The majority of studies aimed to collect data on PA for all seven days of the week [19–21, 23–27, 29, 33]; one study collected data for six [22] and five [32] days, two sampled for four days [28, 30], and two older studies [18, 31] assessed PA over three days. All studies recorded activity during waking hours. Sandberg et al. [28], Larsson et al. [30], and Hedlund et al. [33] additionally utilized a 24-h protocol.

Sampling epoch length of the accelerometer devices differed between the studies [19–28, 30, 31, 33] and ranged from 3 to 60 seconds. Three studies [18, 29, 32] did not report about their devices' sampling epochs at all. Only Voss and colleagues [24] and Hedlund and colleagues [33] reported the sampling frequency (30 Hz) of their devices. Detailed listings of frequency and epoch are displayed in Table 3.

Daily PA in different levels of activity (sedentary, light, moderate, and vigorous) or MVPA were assessed in all studies. However, thresholds, wear-time algorithm and PA intensity classification for different activity levels differed or were not reported at all. Four studies [25, 28–30] did not report how accelerometer data were transferred into PA intensity; in two studies [22, 26] PA could not be determined.

Discussion

Study Quality

The majority of studies were rated “fair” at best with more than a third showing substantial risk for bias with a “poor” rating. Qualitative flaws were mostly found in missing power or effect size, low participation rate or very small cohorts. The lack of key confounding variables in the analysis, no blinding of assessors and an unclear subject selection (Table 1) leaves room for stronger studies in the future. Major statistical and methodological flaws – like reporting inconsistent *p*-values in the abstract versus results section [23] or calculating PA thresholds using an adult recommendation [34] for a cohort of preschool children [18] – should be avoided.

Accelerometer Characteristics

Unless sleep data are relevant, a protocol sampling during waking hours with minimum of four valid data days including one weekend day is recommended [35]. A 7-day protocol is even more advisable, especially in younger participants [36]. Three studies [28, 30, 33] utilized a 24-h recording protocol, the rest sampled just during waking hours. Only 3 separate studies [19, 20, 25] actually adhered to these valid-day inclusion criteria (Table 3).

All but two studies [25, 33] used hip-worn devices. Overall, a possible trade-off between classification accuracy and compliance in hip- versus wrist-worn devices needs to be considered. Ideally studies should be designed with both hip- and wrist-worn devices.

Only two studies reported sampling frequency [24, 33] and none data filters of their devices. Even though exact information tends to be proprietary, it has a major impact on data interpretation and comparability and therefore should be included as detailed as possible in future studies. Highest possible sample-frequency is recommended in multiples of 30 [35].

The younger the subjects, the shorter the epoch length to capture sporadic movements [35]. In light of the WHO recommendation asking for bouts of ≥ 10 min [37], sporadic movements become more negligible, and longer sampling epochs are suitable in adults [35]. All studies in ACHD and young adults reported to adhere to this employing 30–60 s sampling epochs. In pediatric studies epoch length differs (Table 3), and future studies should focus on using the shortest sampling epoch possible. It is recommended to follow the same epoch length and intensity classification criteria which were used in the original calibration study of the respective device. As internal algorithms differ by manufacturer, data processing is device specific. Therefore, it is advisable to use the default intensity classifications. Table 4 contains further recommendations for the application of objective PA assessment in clinical research.

Are Patients with CHD Active Enough?

With a few very specific exceptions, almost all patients with CHD can and should be encouraged by their physician and medical care-takers to achieve general population guidelines for PA [3, 38]. Those are according to the World Health Organization as follows: children and adolescents aged 5–17 should accumulate at least 60 min of MVPA daily with greater benefits above 60 min. Adults aged 18–64 should reach at least 150 min of moderate-intensity aerobic PA throughout the week or do at least

Table 3. PA assessment characteristics

Study	Device	Placement (waterproof)	Assessment length	Data inclusion criteria	Sampling frequency, Hz	Sampling epoch, s
<i>Pediatric CHD (n = 8)</i>						
Kao et al. [18]	RT3	Hip (no)	3 days starting on Friday waking hours	–	–	–
Ewalt et al. [19]	ActiGraph 7164	Hip (no)	7 consecutive days waking hours	≥10 h of data for 3 weekdays and 1 weekend day	–	30
Longmuir et al. [20]	Respironics actical 2.1	Hip (yes)	5 school days and 2 nonschool days waking hours	≥8 h of data on 3 school days and 1 nonschool day	–	15
Stone et al. [21]	ActiGraph GT1M	Hip (no)	7 consecutive days waking hours	≥5 h on ≥4 days incl. 1 weekend day	–	3
Klausen et al. [22]	ActiGraph 77	Hip (no)	6 a.m. – 10 p.m. for 4 weekdays and 2 weekend days at baseline and FU	≥10 h of data on ≥1 weekend day and 1 weekday	–	5
Banks et al. [23]	Respironics actical 2.1	Hip (yes)	5 school days and 2 nonschool days waking hours	≥10 h of data on ≥3 school days and 1 nonschool day	–	15
Voss et al. [24]	ActiGraph GT3X+ and GT9X	Hip (no)	7 consecutive days waking hours	≥600 min/day of data on ≥3 days	30	15
Brudy et al. [25]	Garmin vivofit jr	Wrist (yes)	7 consecutive days waking hours	3 weekdays and 1 weekend day	–	60
<i>ACHD (n = 5)</i>						
Dua et al. [26]	ActiGraph 7164	Hip (no)	7 consecutive days waking hours	–	–	60
Müller et al. [27]	RT3	Hip (no)	7 consecutive days waking hours	–	–	60
Sandberg et al. [28]	ActiHeart	Chest (no)	4 consecutive days 24 h	–	–	30
Opatowsky et al. [29]	ActiGraph GT3X+	Hip (no)	7 consecutive days at baseline and FU waking hours	≥600 min/day of data on ≥3 days	–	–
Larsson et al. [30]	ActiHeart	Chest (no)	4 consecutive days 24 h	–	–	30
<i>Pediatric and ACHD (n = 3)</i>						
Müller et al. [31]	RT3	Hip (no)	3 consecutive days waking hours	–	–	60
Duppen et al. [32]	ActiGraph GT3X	Hip (no)	5 consecutive days waking hours	≥8 h/day on ≥3 days	–	–
Hedlund et al. [33]	ActiGraph GT3X	Wrist (no)	7 consecutive days 24 h	–	30	60
PA, physical activity; CHD, congenital heart disease; ACHD, adults with congenital heart disease.						

75 min of MVPA throughout the week or an equivalent combination of both intensities [37]. Further specific recommendations on indications, type, and intensity for PA and exercise in patients with CHD can be found in international guidelines [4, 39, 40].

Pediatric Patients with CHD

None of the four CSS comparing pediatric patients with CHD to a RC found a difference between patients and healthy in daily PA or MVPA [18, 19, 21, 25]. Only one [25] found their patients reaching the WHO rec-

ommendation for children and adolescents. The remaining three [19, 22, 24] followed quite inactive cohorts in regards to their respective age-related PA recommendations. The cohort of Stone et al. [21] was also quite inactive but was compared to the Canadian PA Guidelines for the Early Years [41] with some of their patients being older than the used institutional recommendation.

Two CSS without RC evaluated PA in regard to CHD severity or CHD subgroups and did not find differences in mean MVPA [23, 24], while others [25] reported com-

Table 4. Recommendations for the application of PA assessment in clinical research

Sampling protocol	7-day sampling period; ≥ 4 valid days including 1 weekend day capturing all waking hours; clear data inclusion criteria defined
Device placement	Possible trade-off between hip (better for intensity classification accuracy) & wrist-worn (better for wear-time compliance) devices to consider; ideally both in research settings
Sampling frequency	Highest possible in multiples of 30
Epoch length	Shorter (3–10 s) in children & adolescents to capture sporadic movement; longer (30–60 s) in adults
Intensity classification & data filters	Follow initial calibration study guidelines or default settings of the device
PA guidelines/reference	Age & region/country-appropriate guidelines must be employed; preferably WHO criteria and country specific guidelines; own recruited healthy reference cohort is advisable
Confounders to consider	Age, sex, BMI, CHD severity, weather, and seasonality
PA, physical activity.	

plex CHD to be associated with not meeting the WHO recommendation on PA.

While two studies [24, 25] report a decrease in daily PA with increasing age, Longmuir et al. [20] were able to keep MVPA constant through an intervention study over 22 months of long-term FU contrasting the expected decrease in daily PA with age. Furthermore, all three studies dealing with pediatric and ACHD (but generally rather young cohorts) found an age-related decrease in PA [31–33].

Even though the influence of weather and seasonality on PA behavior has been demonstrated [42], only a few studies controlled their results [20, 24, 25] therefor. One [20] found an association between spring season and PA in pediatric patients with CHD.

Pediatric and ACHD

Müller and co-workers [31] found 72% of patients with total cavopulmonary connection to reach ≥ 60 min activity ≥ 3 MET on ≥ 5 days per week [43]. Similarly, Duppen et al. [32] had 70% of patients meeting ≥ 60 MVPA min/day at baseline before heading into intervention. Young male patients were significantly more active than female which was also confirmed in ACHD [28]. However, Duppen et al. [32] did not improve daily PA or percentage of MVPA time through 12 weeks of aerobic training in an already active young cohort. In contrast, Hedlund et al. [33] showed 90% of their young population to reach ≤ 60 MVPA min/day, an overall similar measured MVPA for patients and age-and-sex-matched RC,

and an age-related MVPA decrease. Furthermore, patients spent less time sedentary and more time in light activity than RC [33].

ACHD

One of the three CSS [27] reported 76% of ACHD to be fairly active reaching ≥ 30 MVPA min/day, while the other two [28, 30] found no differences in MVPA against a healthy RC and in the proportion that reached the WHO guideline [37]. The latter two also found about half of all ACHD and RC not reaching daily PA recommendations. It is important to note that both Sandberg et al. [28] and Larsson et al. [30] calculate the WHO recommendation as 21.4 MVPA min/day. However, the WHO recommends to split the ≥ 30 MVPA min/day in bouts of ≥ 10 min [37]. Therefore, these results need to be interpreted carefully.

The clinical controlled trial [26] and the RCT [29] conducted in ACHD did not use a healthy RC. Dua and colleagues [26] increased the adherence to the UK National Guidelines for PA [44] from 14 to 56% and mean MVPA minutes almost doubled after 10 weeks intervention. Furthermore, patients with higher NYHA class were less active at baseline and more able to improve their PA through training. Contrarily, Opatowsky and colleagues [29] found only a trend toward more MVPA time, and no difference regarding PA at baseline and FU after 12 week cardiac rehabilitation intervention versus standard of care.

Generally, it seems like the older the cohorts, the less active CHD patients are both against healthy RC and in-

stitutional PA recommendations. Few of the herein mentioned interventional studies tried to improve PA showing mixed results on the efficacy of these interventions. Large studies with a focus on CHD subgroups currently do not exist.

PA and Exercise Capacity

Eight studies determined parameters for physical capacity, mostly through cardiopulmonary exercise testing in CHD patients, but four reported cardiopulmonary exercise testing outcomes separately [20, 22, 29, 32], while one [26] only examined maximal walking time without associating PA. Müller and colleagues [27] found a moderate relationship between daily activity and exercise capacity and PA to be reduced in patients with lower VO_{2peak} [31], while Banks et al. [23] found only a trend in the association between MVPA and higher percent-predicted VO_2 . All 3 studies did not have a healthy RC.

Considering the importance of exercise capacity in predicting mortality in CHD patients [45], future studies should explore the association to PA in this population more in detail. In a similar physiological context, there is no information on the relationship between daily PA and cardiovascular risk in CHD patients. Exploring this triumvirate of objectively measured PA, exercise capacity, and cardiovascular risk holds potential for further studies and eventually improving medical aftercare in CHD patients.

PA and Quality of Life

Müller et al. [27] found only a poor correlation between daily PA and several QoL domains in ACHD. In pediatric patients with CHD, Banks and co-workers [23] showed higher MVPA to be associated with higher self-efficacy in pediatric patients with CHD, while Müller and colleagues [31] found QoL to be related to daily activity in children with CHD younger than 14 years. Duppen et al. [32] confirmed weak correlation between self-estimated QoL and daily PA in adolescents and young ACHD. Hedlund and co-workers [33] sampled but did not analyze PA in relation to QoL. Therefore, clear answer on the relationship between objective PA and QoL in CHD patients can currently not be given leaving room for future, large scale studies.

Limitations

Sample size of the majority of included studies was generally small. Comparison between studies presented difficulties because device types and characteristics, data

classification criteria, and the guidelines PA was measured against differed substantially.

The majority of studies were cross-sectional, five were interventional. The main goal of this review was to compare the status quo by evaluating quality of methods, outcome measures, and results and not to assess interventions of the separate studies. Therefore, we decided to critically appraise all articles according to the NHLBI Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies. We acknowledge that to evaluate the intervention quality the NHLBI Quality Assessment of Controlled Intervention Studies is the appropriate tool for RCT or clinical controlled trial. However, both questionnaires contain similar key methodological items.

Conclusion and Clinical Recommendation

The methodology and assessment of PA was fair at best in most of the studies. Due to methodological differences of the studies, no clear answer on how active CHD patients really are can currently be given.

Objective PA measures are crucial in identifying patients at risk for inactivity. Therefore, larger studies adhering to consistent measuring and reporting standards exploring PA in CHD patients, also in the context of different angles such as exercise capacity or QoL, are needed. These studies should employ their own healthy RC and compare PA against institutional guidelines. Future studies need to consider device placement, sampling protocol and frequency, epoch length, intensity classification thresholds, valid-day inclusion criteria, and wear-time definition more carefully.

Conflict of Interest Statement

No conflict of interest. No competing interests for any of the authors.

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Author Contributions

J.M. was responsible for conception of the review and further monitored the search process and quality assessment of applicable studies. L.B. and M.M. screened and assessed the search results. L.B. also developed the search strategy and drafted the manuscript.

L.G.C. was the third reviewer to dissolve in case of disagreement between the first and the second reviewer and further gave valuable feedback for improving the manuscript. R.O., A.H., and P.E. gave important input for revising and improving the quality of the manuscript. All authors have read and approved the final version of the manuscript.

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