



Article

The effects of aquatic exercises on body functions, activity and participation in children and adolescents with cerebral palsy: a systematic review

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ABSTRACT:

Introduction: Aquatic exercises may benefit children and adolescents with cerebral palsy (CP). Nevertheless, there are few studies about the efficacy and safety of those interventions for this population. Therefore, the objective of this study was to perform a systematic review of the effects of aquatic interventions in children and adolescents with CP, considering outcomes related to the body structure, function, activity, and participation; based on the World Health Organization International Classification of Functioning, Disability and Health framework (ICF).

Methods: We searched for experimental or quasi-experimental studies about aquatic exercises' effects on children and adolescents with CP. We used the databases PubMed/MEDLINE, CINAHL, Scielo, LILACS, SPORTDiscus, Cochrane Reviews, Trials, and PEDro, from January 1st, 2011, to December 31st, 2020. In addition, the risk of bias was assessed with the PEDro tool.

Results: 10 randomized clinical trials and five quasi-experimental studies were included. The focus was on aerobic activities in children with spastic CP, mainly classified as levels I to III in the Gross Motor Function Classification System (GMFCS), outcomes related to structure and function, and activity. Thirteen studies reported positive effects, but the methodological quality was low.

Discussion: Aquatic exercise programs are feasible and demonstrate a positive effect on this population. There is a need for studies of better methodological quality, including children classified as GMFCS V and outcomes based on the domain of participation.

Keywords: cerebral palsy; water; aquatic; hydrotherapy; swimming

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Introduction

Cerebral palsy (CP) is the most common physical disability in childhood, with an estimated prevalence of 2.1 cases per 1000 live births in developed countries (Oskoui, 2013). The definition of CP comprises a group of non-progressive, yet permanent, disorders affecting the fetal or infant brain and impacting movement and posture, with activity limitation (Rosenbaum, 2007). Although the signs may be present in the first months of life, the diagnosis is typically established between 12 and 24 months of age (Hubermann, 2016). The etiological factors may vary depending on the gestational age group and clinical CP subtype. Still, they usually include risk factors present during pregnancy, birth, and the postneonatal period, as well as genetic predisposition (Nelson, 2008). Individuals with CP can be classified according to their gross motor function with the Gross Motor Function Classification System (GMFCS), a 5-level ordinal system. Level I represents the less severe gross motor impairment, and group V is the most severe (Palisano, 1997).

Usually, an impact on motor function is present in all children with CP (Rosenbaum, 2007). Nevertheless, their motor impairments may be complex and variable, including abnormal muscle tone, balance, and coordination; reduced muscle strength; decreased selective motor control; and secondary musculoskeletal abnormalities, such as muscle contractures and joint deformities (Papavasiliou, 2009). Furthermore, children with CP have higher levels of a sedentary lifestyle, and their prevalence of obesity has been increasing over time, under the trend observed in the general pediatric population (Palisano, 1997). Those factors underscore the importance of designing specific physical activity programs for this population to improve motor function and control weight gain (Rogozinski, 2007; van den Berg-Emons, 1995).

The fitness of children with disabilities has been emphasized as a core component of the ‘F-words’ (function, family, fitness, fun, friends, and future), a framework based on the International Classification of Functioning, Disability and Health (ICF) from the World Health Organization (WHO) (WHO, 2001) to plan services, research, and assistance to this population (Rosenbaum, 2012). Aquatic physical activity is appealing and attractive for children with CP because it is engaging and fun (Kelly, 2005). Furthermore, it may help to improve their muscle strength, cardiovascular function, and gross motor skill performance, keeping their physical fitness (Kelly, 2005). In addition, the water’s buoyancy helps decrease joint loading and impact, potentially reducing the negative effect of poor balance and difficult postural control (Kelly, 2005). Nevertheless, there is a paucity of studies on the safety and

potential benefits of aquatic activities in this population (Kelly, 2005).

In 2011, a systematic review summarized the available body of evidence regarding the effects of aquatic exercise programs in the population of children and adolescents with CP, considering outcomes based on the ICF. The authors found that most of the research was focused on higher-functioning children with CP, with a substantial heterogeneity of interventions and outcome measures, as well as a low methodological quality of the studies (Gorter, 2011). After that, no other reviews aimed at summarizing the evidence about aquatic exercises as interventions for this specific population using an ICF-based outcome evaluation approach. We considered it necessary to revisit this research question 10 years after the last review to update the current evidence and inform future research. Therefore, we conducted a systematic review of the literature to evaluate the effects of aquatic activities programs in children and adolescents with CP on the domains of body structure and function, activity, and participation of the ICF from the WHO.

Materials and Methods

This systematic review followed the Preferred reporting items for systematic review and meta-analysis protocols – PRISMA guidelines (Page, 2020).

Search strategy

We searched the databases Pubmed/MEDLINE, CINAHL, Scielo, LILACS, SPORTDiscus, Cochrane, Trials, and PEDro, from January 1st, 2011, to December 31st, 2020, to retrieve experimental or quasi-experimental studies reporting outcomes of children with CP after aquatic interventions, based on the ICF framework, from the WHO.

The following keywords were used: “cerebral palsy” AND “hydrotherapy” OR “swimming” OR “aquatic” OR “water.” The search was performed through the boolean operators “AND” and “OR” with a combination of descriptors. The first selection was based on the titles, the second based on the abstracts, and the final on the full text.

The study was registered at the International Prospective Register of Systematic Reviews (PROSPERO), registration number CRD42021231425. There were no deviations from the protocol.

Study inclusion criteria

We included experimental or quasi-experimental studies (i.e., randomized clinical trials, intervention studies without randomization or control group) that included children and adolescents (aged 1 to 21 years)

with a diagnosis of CP (at least 1 participant), GMFCS levels I to V, subjected to interventions involving aquatic exercises, including aerobic, anaerobic, strength or other types of activities performed in the water. If present, the control group could not have received activities in the water environment.

We considered the definitions of interest, based on the review undertaken by Gorter et al., as follows: aerobic was considered as any activity to enhance cardiorespiratory fitness, usually performed for an extended period, including water walking and swimming lengths; anaerobic activities were considered as activities with short lasting and higher intensity, including jumping; strength-based activities included those involving aquatic resistive training to increase muscle strength; other exercises comprised activities that did not fill the criteria for the different categories, such as stretching and aquatic play (Gorter, 2011).

Outcome measures were classified according to the ICF framework: body function, activity, and participation. Body function was represented by energy expenditure index (EEI), muscle strength measures, range of motion, gait velocity, ventilatory and metabolic measurements. The activity domain comprised measures such as the Gross Motor Function Measure (GMFM), the functional reach test, the 6-minute walk test (6MWT), and timed up and go (TUG). Finally, the participation domain included measures such as the Canadian Occupational Performance Measure (COPM) and self-perception scales.

The exclusion criteria included: studies not reported in English or Portuguese, studies without primary data, repeated reports of the same setting, or papers without appropriate reporting to allow the data extraction.

Two authors (AC and JRF) independently screened the titles, abstracts, and full texts and selected the studies according to the inclusion criteria. In cases of disagreement, the opinion of a third reviewer (JRA) was requested to reach a consensus.

Data extraction

The data extraction was performed through a standard form. Regarding the study features, we extracted data about: the title, authors, country of origin of the children, year of publication, Journal, aims, and study design. About the participants, we collected the sample size, age range, CP subtype, distribution of motor impairment, and GMFCS level. Regarding the intervention features, we retrieved information about the type of intervention (classified as aerobic, anaerobic, strength, or others), session duration, session length, session frequency, and characteristics of the control group. Finally, regarding the outcomes, we extracted

the measures and instruments used, their classification according to the ICF domain, and a summary of the main results.

After the data extraction, the studies that met the inclusion criteria were included in a qualitative analysis.

Risk-of-bias assessment

The studies were evaluated according to the risk of bias through the PEDro scale. Using the PEDro tool (de Morton, 2009), we assessed each selected study regarding all the criteria, awarding points if they were satisfied by the report's analysis. The final score considered the sum of all awarded points for each study, not considering criterion 1, following the instrument guidance, because this evaluates the study's generalizability. Finally, the mean points of the final score were calculated.

Results

Study selection and methodological quality

The flowchart with the selection process of the articles is shown in Figure 1. Our search strategy yielded 112 articles. After removing duplicates and exclusions for other reasons, 100 titles and abstracts were screened, of which 54 were excluded because they did not fulfill the inclusion criteria.

After assessing 46 full texts for eligibility, 31 papers were excluded (reasons provided in the flowchart). The remaining 15 papers were included in the detailed analysis (details summarized in Table 1) (Adar, 2017; Akinola, 2019; de Araújo, 2018; Ballaz, 2011; Ballington, 2018; Jorgic, 2014; Declerck, 2013; Declerck, 2016; Depiazzi, 2021; Dimitrijevic, 2012; O'Sullivan, 2011; Fragala-Pinkham, 2014; Lai, 2015; Morgan, 2012; Sharan, 2018).

As for the risk of bias assessment, the included studies did not address most of the checklist items, which shows the overall poor methodological quality of the reported data. The risk of bias analysis is described in Supplementary Tables 1 and 2. The mean scored points in the PEDro Checklist was 4.2 out of 10 total possible points.

Studies feature

The studies were published between 2011 and 2021. Most studies were from the United Kingdom, the United States, and Serbia, with two studies from each country. The other papers were from Turkey, Nigeria, Brazil, Canada, South Africa, Australia, Ireland, Taiwan, and India. Ten studies were randomized control trials, nine using a parallel design and one with a cross-over design.

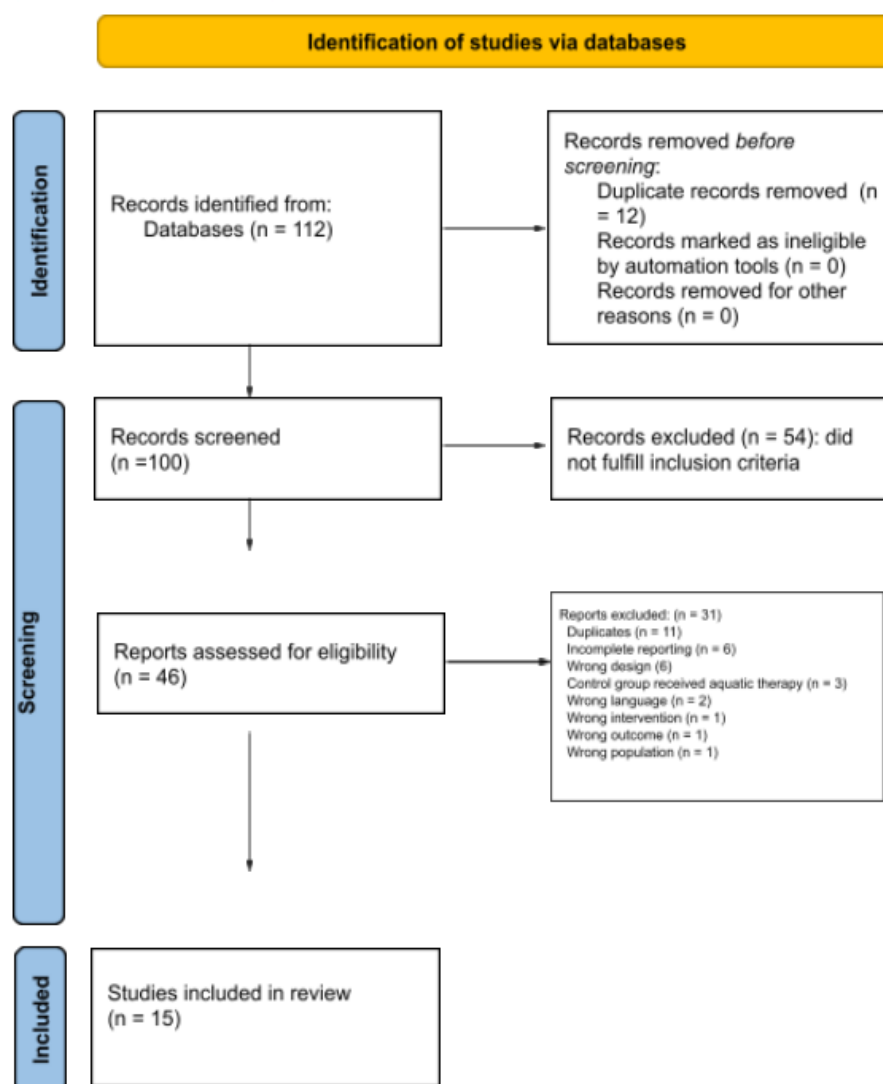


Figure 1. PRISMA flow diagram.

Five studies used a quasi-experimental approach, either with a single group pre-post design or a non-randomized one.

Sample characteristics and outcomes

The 15 studies included an overall sample of 294 children, with a minimum of 8 participants and a maximum of 40 participants per study. Most of the studies had school-aged children (mean age 10.4 years). All the studies included children with spastic CP. Only three studies were undertaken with dyskinetic participants and included only one child with ataxia. The majority included children in levels I to III of GMFCS. Only two were conducted with participants in level V of GMFCS, with just 7 participants in this level.

The intervention was mainly focused on aerobic activities, which was applied in all the studies, although the majority described that they were somewhat individualized (number of repetitions, intensity,

progression). The number of sessions varied from 6 to 36, ranging from 35 to 100 minutes for each session, 2 to 6 times per week, from 6 to 14 weeks. The control group either was absent or received conventional land-based therapies. The studies did not report the outcome analysis based on the type of activity (aerobic, anaerobic, strength, or others).

As shown in Table 1, all three domains of body structure and function, activity, and participation of the ICF were represented in outcomes evaluated in six studies. The most evaluated domain was activity, represented in fourteen studies, followed by body structure and function in eleven studies. Participation was evaluated in only six studies.

Only two studies did not show significant differences in the outcomes among the groups. However, the risk of bias assessment showed a low overall methodological quality due mainly to the absence of randomization, blinding, or data reporting.

First author, country, year	Sample characteristics	Interventions	Outcomes	Results
Adar et al. Turkey, 2017.	N = 32 Mean age of 9.7 (SD 2.7) years. Spastic CP, hemiplegia or diplegia. GMFCS I-IV	<u>Intervention:</u> aerobic and strength exercises; 30 sessions of 60 minutes, 5 times per week, for 6 weeks. <u>Control:</u> land-based activities.	<u>Structure and Function:</u> ultrasonographic assessment of spastic gastrocnemius muscle, modified Ashworth scale. <u>Activity:</u> TUG, GMFM 88, WeeFIM. <u>Participation:</u> Pediatric Quality of Life Inventory (PedsQL)-CP Module.	No differences in the functional outcomes.
Akinola et al. Nigeria, 2019.	N = 30 Age ranges from 1 to 12 years. Spastic CP. GMFCS II-V	<u>Intervention:</u> aerobic; 20 sessions; duration of 100 minutes, 2 times per week, for 10 weeks. <u>Control:</u> same activities, but land-based.	<u>Activity:</u> GMFM 88.	Only the experimental group significantly improved all dimensions of gross motor function except for walking, running, and jumping.
Araújo et al. Brazil, 2018.	N = 16 Mean age of 11.7 (SD 2.5) years. Spastic CP GMFCS II and III	<u>Intervention:</u> aerobic, strength, and others; 16 sessions; duration of 35 minutes, 2 times per week, for 8 weeks. <u>Control:</u> conventional therapies.	<u>Structure and Function:</u> EMG, VM, IGE, Wells' Fluxmeter. <u>Activity:</u> TCMS, GMGM-88, 6MWT, TUG. <u>Participation:</u> CHQ PF-50, gait EVA.	The intervention group had better dynamic trunk balance and balance reaction.
Ballaz et al. Canada, 2011.	N = 12 Mean age of 17.9 (13-21) years. Spastic and dyskinetic CP. GMFCS I to IV	<u>Intervention:</u> aerobic and others; 20 sessions; duration of 45 minutes, 2 times per week, for 10 weeks. <u>Control:</u> absent.	<u>Structure and Function:</u> EEI; gait analysis; isometric strength testing on quadriceps and hamstrings. <u>Activity:</u> GMFM.	The program improved gait efficiency, as measured by the EEI.
Ballington et al. South Africa, 2018.	N = 10 Mean age of 11.0 (SD 0.08) years Spastic CP GMFCS I to III	<u>Intervention:</u> aerobic, anaerobic, and others; 16 sessions of 30 minutes, 2 times per week, for 8 weeks. <u>Control:</u> no aquatic activities. A wash-out period of 1 month.	<u>Activity:</u> GMFM 66.	The aquatic program had an increased motor function after the intervention compared to the control group.
Jorgic et al. Servia, 2014.	N = 15 Mean age of 12.26 (SD 2.97) years. Spastic and dyskinetic CP. GMFCS I to III	<u>Intervention:</u> aerobic, strength, and others; 36 sessions; duration of 60 minutes, 3 times per week, for 12 weeks. <u>Control:</u> absent.	<u>Structure and Function:</u> Range of motion measurements.	After the experimental program, a statistically significant increase was observed in the range of motion for flexion and abduction at the shoulder joint.

First author, country, year	Sample characteristics	Interventions	Outcomes	Results
Declerck et al. UK, 2016.	N = 14 Mean age of 12.26 (SD 2.97) years. Spastic CP. GMFCS I to III	<u>Intervention:</u> aerobic, anaerobic, and others; 20 sessions; duration of 40-50 minutes, 2 times per week, for 10 weeks. <u>Control:</u> usual physical therapy program without scheduled swimming.	<u>Structure and Function:</u> Faces Pain Scale-Revised, Visual Analogue Scale, PedsQL Fatigue. <u>Activity:</u> 1-minute fast walk test; WOTA 2. <u>Participation:</u> level of satisfaction with the program on a 5-point Likert scale.	High levels of enjoyment were observed among participants in the intervention group; in the intervention group, improvements were observed in the swimming skills without adverse effects on pain or fatigue.
Declerck et al. UK, 2013	N = 8 Median age of 10.2 (IQR 2.3) years. Spastic, dyskinetic, ataxic CP. GMFCS I to III	<u>Intervention:</u> aerobic, anaerobic, and others; 12 sessions; duration of 60 minutes, 2 times per week, for 6 weeks. <u>Control:</u> none.	<u>Structure and Function:</u> Hand grip strength with the Jamar Hydraulic Hand Dynamometer. <u>Activity:</u> Jebsen-Taylor test of hand function, GMFM-88, 10-MWT, WOTA2. <u>Participation:</u> CP Quality of Life Questionnaire for Children, Assessment of Life Habits Short Form questionnaire.	There were improvements in adjustment and functioning in water.
Depiazzi et al. Australia, 2021.	N = 12 Mean age of 14.5 (SD 2.0) years. Spastic CP GMFCS II	<u>Intervention:</u> aerobic; 20 sessions; duration of 40 minutes, 2 times per week, for 10 weeks. <u>Control:</u> conventional therapy.	<u>Structure and Function:</u> Cardiopulmonary exercise testing, dual-energy X-ray absorptiometry scan, Bath Adolescent Pain Questionnaire-5, Modified Brief Pain Inventory. <u>Activity:</u> Fatigue and physical activity modules of the Pediatric Quality of Life Inventory (PedsQLTMv4.0) – adolescent version. <u>Participation:</u> Canadian Occupational Performance Measure.	This aquatic high-intensity interval training protocol was feasible, safe, and well tolerated in adolescents with cerebral palsy GMFCS II.
Dimitrijevic et al. Serbia, 2012	N = 29 Age ranges from 5 to 14 years. Spastic CP GMFCS I-V	<u>Intervention:</u> aerobic, anaerobic, and others; 12 sessions; duration of 55 minutes, 2 times per week, for 6 weeks. <u>Control:</u> other activities.	<u>Structure and Function:</u> GMFM 88 and WOTA 2.	There were significant effects after the intervention on the gross motor function and water skills of children with CP.
O'Sullivan et al. Ireland, 2011.	N = 12 Age ranges from 4 to 14 years. Spastic CP GMFCS I to III.	<u>Intervention:</u> aerobic, strength, and others; 6 sessions; duration of 45 minutes, 1 time per week, for 6 weeks. <u>Control:</u> land activities.	<u>Structure and Function:</u> handheld dynamometer. <u>Activity:</u> GMFM.	Hydrotherapy provided no additional benefit.

First author, country, year	Sample characteristics	Interventions	Outcomes	Results
Fragala et al. USA, 2013	N = 8 Median age of 10.6 (SD 3.5) years. Spastic CP GMFCS I to III	<u>Intervention:</u> aerobic, anaerobic, strength, and others; 28 sessions; duration of 60 minutes, 2 times per week, for 14 weeks <u>Control:</u> none	<u>Structure and Function:</u> The Brockport modified curl-up, the Brockport isometric push-up, lateral step-ups, shuttle run test, and Pediatric Berg Balance Scale. <u>Activity:</u> GMFM, 6MWT.	Significant improvement was observed for the primary outcomes (gross motor function and walking endurance).
Lai et al. Taiwan, 2015.	N = 24 Mean age of 91.5 (SD 36.5) months. Spastic CP. GMFCS I to IV	<u>Intervention:</u> aerobic, anaerobic, strength, and others; 24 sessions; duration of 40 minutes, 2 times per week, for 12 weeks. <u>Control:</u> land activities.	<u>Structure and Function:</u> Modified Ashworth Scale. <u>Activity:</u> GMFM 66. <u>Participation:</u> Physical Activity Enjoyment Scale score, Vineland Adaptive Behavior Scale, Cerebral Palsy Quality-of-Life–parent proxy scale.	The pediatric aquatic therapy group significantly improved gross motor function and enjoyment levels.
Morgan et al. USA, 2012.	N = 32 Mean age of 10.5 (SD 3.9) years. Spastic CP. GMFCS I and II	<u>Intervention:</u> aerobic and anaerobic; 30 sessions; variable duration, 3 times per week, for 10 weeks. <u>Control:</u> conventional activities.	<u>Structure and Function:</u> Preferred walking speed (PWS), maximal walking speed (MWS). <u>Activity:</u> 5-minute walk distance (5MWD).	Statistically significant and functional improvements in measures of walking performance were observed after the intervention.
Sharan et al; India; 2018	N = 40 Mean age of 7,3 (SD 3.0) years	<u>Intervention:</u> details not described; 36 sessions; variable duration, 6 times per week, for 6 weeks. <u>Control:</u> land activities.	<u>Activity:</u> Quality of Upper Extremity Skills Test (QUEST) and Melbourne Assessment of Unilateral Upper Limb Function (MAUULF).	The intervention group showed better results in the primary outcomes.

TUG: Timed Up and Go Test; **GMFM-88:** Gross Motor Function Measure-88; **WeeFIM:** Wee Functional Independence Measure; **TCMS:** Trunk Control Measurement Scale; **EMG:** surface electromyography; **6MWT:** six-minute walk test; **VM:** mean walking speed; **IGE:** energy expenditure index; **EVA:** visual analog scale; **CHQ PF-50:** Child Health Questionnaire; **WOTA 2:** Water Orientation Test Alyn 2; **10-MWT:** 10-meter walking speed.

Table 1. Main results of the included articles in the systematic review

Discussion

The present study was a systematic review summarizing the current evidence on the effects of aquatic activities as interventions for children and adolescents

with CP to improve outcomes focused on the ICF domains over the last 10 years. We found that the focus of the studies continued to be on aerobic activities in children with high-functioning spastic CP, mainly

related to body structure, function, and activity. Most studies reported positive effects of the interventions, but the overall methodological quality was low.

Similar to our findings, the evidence up until 2011, summarized by Gorter et al. (Gorter, 2011), also showed a predominance of participants with spastic CP in higher levels of the GMFCS, focused on aerobic interventions in water, with heterogeneous protocols and mainly directed to structure, function, and activity domains. On the other hand, in the last ten years, we observed a trend to include larger sample sizes, and the more frequent use of randomization and control groups, even though the overall methodological quality is still low. Furthermore, compared to the 2011 systematic review, we showed a more considerable number of studies reporting outcomes focused on participation: 6 studies, compared to only two studies, reported by Gorter et al. (Gorter, 2011).

In another systematic review (Roostaei, 2017) of the effects of aquatic intervention in children with CP, but focused on gross motor skills as outcomes, the authors found similar patterns in terms of good feasibility and safety of the interventions, but with poor methodological quality, small sample sizes and a predominance of participants in higher levels of GMFCS, consistent with our findings.

Most studies may not have included children classified as GMFCS levels IV and V because of the higher prevalence of comorbidities and potential difficulties with regular follow-up, considering clinical issues that may hinder their participation. This brings additional challenges for this subset of the CP population, especially considering they may already have more limited treatment options to stay physically active. One possible solution could be to innovate ways to assess the outcomes, especially related to the participation domain, including quality of life evaluation through telehealth strategies, which could facilitate follow-up and reduce the need to go to the research facility.

Regarding the intervention, we noted a lack of consistency in the treatment protocols among the studies, with heterogeneity regarding the type of activity, length,

and frequency. Therefore, it would be interesting for future research focuses on applying standardized protocols that could be comparable in different settings.

As strengths of the present study, we emphasize the methodological aspects, including the systematic review database registration, the use of a focused research question and search strategy, with the use of multiple relevant databases, and the assessment of outcomes based on the ICF, which allows a meaningful evaluation of the current scope of the literature on this theme. Nevertheless, we also acknowledge some important limitations, including language restrictions, since articles not published in English or Portuguese were excluded. However, publications from other non-English speaking countries reported in English were retrieved, as well as the possibility of publication bias, considering that studies with negative results are less likely to get published. Furthermore, the characteristics of the interventions described in the studies were heterogeneous, which may hinder a clear conclusion about their effects when the data are analyzed collectively.

Our results show that there still exists a gap in the literature regarding the long-term efficacy and safety of aquatic physical activity in children and adolescents with CP based on ICF outcomes. Therefore, there is a pressing need for future research with higher methodological quality (larger sample sizes, appropriate pre-planned sample size calculations, proper randomization, and outcome assessors blinding), the inclusion of participants with other types of CP and levels of functioning, especially GMFCS levels IV and V, as well as outcomes focused on meaningful and gold-standard participation outcomes. Participants with higher GMFCS levels should be assessed separately in studies designed to evaluate the efficacy and safety of those interventions in this population.

Conclusion

This systematic review demonstrated that aquatic activity programs have a positive effect on the population of children and adolescents with CP, but the evidence had low methodological quality, the studies included mainly children of higher motor functioning and

focused mostly on the domains of body structure, function, and activity. Those results underscore the need for future studies of better methodological quality, such as proper randomization, blinding of outcome assessors, the inclusion of children classified as GMFCS V, and outcomes based on the domain of participation, which may be more relevant and meaningful for individual patients and their families.

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Conflicts of Interest: The authors declare no conflict of interest.

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