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Clinical Biomechanics 100 (2022) 105817

Contents lists available at ScienceDirect

Clinical Biomechanics

journal homepage: www.elsevier.com/locate/clinbiomech

Running capacity in children with bilateral cerebral palsy: What are the biomechanical and neuromotor differences between runners and walkers?

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ARTICLE INFO

Keywords:
Cerebral palsy
Running
Muscle strength
Spasticity
Selectivity
Physical examination

ABSTRACT

Background: Running is a high-level locomotor activity requiring more from joints, muscles and a more complex interaction of the neuromuscular system than walking. High-level locomotor activity has the potential to shed light on motor function deficits that lower-level activity does not reveal. Therefore, the study aimed to compare biomechanical and neuromotor function between a group of children with bilateral cerebral palsy who are able and unable to run.
Methods: Retrospectively, children with bilateral cerebral palsy aged between 6 and 18 years who completed a clinical gait analysis between 2006 and 2019 were included. Participants were categorized as walkers or runners based on the presence of a double floating phase. Spasticity, selectivity, muscle weakness, and passive range of motion of the lower limbs were measured and dichotomized as «normal» or «abnormal» based on reference values. Functional tasks reflecting balance (standing on one leg) and power (single leg and two-legged jumps) were realized and evaluated as failure or success.
Findings: 75 children with bilateral cerebral palsy (53 runners/22 walkers) were included. Children classified as runners were stronger (hip flexors, $p = 0.006$; hip abductors, $p = 0.022$; knee flexors, $p = 0.001$; dorsiflexors, $p = 0.014$), had greater selectivity (hip flexors, $p = 0.011$; dorsiflexors, $p = 0.001$; plantiflexors, $p = 0.043$) and lower spasticity at the knee extensors ($p = 0.045$). No differences were observed in the passive range of motion between the two groups. Children classified as runners performed better at all tasks of balance and power ($p < 0.05$).
Interpretation: Flexors muscles strength and selectivity and knee extensor spasticity are key points for running ability in children with bilateral cerebral palsy.

1. Introduction

Physical activities, including running, are important for general health by preventing chronic illnesses and it can contribute, in youth with cerebral palsy (CP), to maintain or improve motor function as well as general participation in everyday life. However, in children with CP, physical activity limitations are reported including running capacity and performance (Rimmer, 2001). Running capacity refers to an individual ability to execute a running pattern, which can be defined as taking quick steps so that both feet are simultaneously off the ground (World Health Organization, 2007). On the other hand, running performance refers to what an individual does in life situations (World Health Organization, 2007). Although some children with CP can run, this capacity is limited for many individuals and more specifically among children with bilateral CP (BCP) (Böhm et al., 2018).

Running is a high-level locomotor activity requiring greater mobility of joints, muscle power, and control of voluntary movement than walking. To move the body forward during running in typically developed (TD) individuals, higher loads on the musculoskeletal systems are required (Arampatzis et al., 1999).

While running, greater muscle strength, generated faster is required to propel the body forward (Dorn et al., 2012). Previous studies suggest that muscle weakness would be a limiting factor for motor skills and the capacity to increase walking or running speed in children with CP

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https://doi.org/10.1016/j.clinbiomech.2022.105817
Received 14 February 2022; Accepted 8 November 2022
Available online 11 November 2022

(Beckman et al., 2016; Pouliot-Laforte et al., 2020). More specifically, a reduced power generation at the ankle has been observed in children with CP compared to TD children while walking, yet similar power generation at the hip was observed while running (Davids et al., 1998). Knowing that running requires greater velocity, spasticity could also interfere with running activity. In children with CP, quadriceps spasticity is associated with a decreased angular knee flexion velocity, knee peak flexion, gait velocity, and stride length while walking (Damiano et al., 2006). Impaired selective voluntary motor control interferes with the ability to control force, speed, and timing of muscle contractions, disturbing the pattern of voluntary movement (Sanger et al., 2006) and therefore, could limit the capacity to run. Children with CP can experience muscle contractures limiting joints range of motion (RoM) (McDowell et al., 2012). While running, they tend to increase their joint RoM in the sagittal plane compared to walking (Chappell et al., 2019a; Davids et al., 1998). Indeed, running involved an increase in dynamic RoM in ankle peak dorsiflexion and hip and knee peak flexion (Dugan and Bhat, 2005). Therefore, the impact of restricted RoM on running capacity is unclear. Running patterns lead to an increase in step length and a decrease in step duration which increases instability. Wider steps are observed while running in children with CP suggesting an adaptation to impair postural control (Rethwilm et al., 2021).

Despite few studies comparing running performance between children with CP and TD children, little is known about the running capacity and the biomechanical and neuromotor differences between children with CP who can and cannot run. One previous study from Böhm et al. (Böhm et al., 2018), identified contributing factors of running capacity by comparing clinical measures between runners and non-runners. They observed, in a group comprising unilateral and bilateral forms of CP with Gross Motor Function Classification System (GMFCS) level II, higher performance on a single leg jump and single-leg stance in the runner group compared to the walker group. They also observed a lower body mass index (BMI) and spasticity rate of the rectus femoris in the runner group, but similar muscle strength (Böhm et al., 2018). However, no information on selective motor control and RoM was reported. Including those measures would give a more detailed portrait of the determinants of running capacity. Moreover, separating the topographical classification of CP, i.e. unilateral or bilateral, could also strengthen the analysis.

To add knowledge about running capacity, the present study aimed to compare biomechanical and neuromotor function, measured in clinical assessment, between children with BCP who can and cannot run. This objective will help to define a clinical portrait of runners in children with BCP. Running capacity assessment has the potential not only to quantify a more complex motor skill but also to better identify motor function deficits in this population. Determining the key parameters of the capacity to run would allow better targeting of interventions, more specific recommendations, and potentially improve or maintain higher motor function in youth with BCP.

2. Materials and methods

Retrospectively, participants were selected from the database of children who had undergone a clinical gait analysis (CGA) in a tertiary hospital (Kinesiology Laboratory of Geneva University Hospitals, Switzerland) between 2006 and 2019. From the database, all children who fulfilled the following inclusion criteria were included: a diagnosis of spastic BCP, aged between 6 and 18 years old, able to walk independently without assistive devices, and able to follow simple verbal instructions. Exclusion criteria were surgical intervention 12 months before the gait analysis and Botulinum Toxin injection six months prior. In case of multiple visits from the same individual, the first visit meeting the inclusion and exclusion criteria was chosen. The study was approved by the local ethics committee (CER no. 2018–00229). Informed consent was obtained from all participants and their respective legal guardians since the approval (March 2018). For CGA performed before this date, a consent exemption was granted by the local ethics committee.

During the CGA, participants were instructed to run barefoot at comfortable self-selected speed along a 10 m walkway. One way along the walkway was considered as a trial and participants were asked to complete at least four trials with a small pause between each trial. A video camera, positioned in the middle of the walkway, recorded the trials. At least four gait cycles were visible on each video recording of a trial. The video recordings were used to classify participants. Based on the presence of a double floating phase, which defined a running pattern (Davids et al., 1998), participants were classified as *runners*. To ensure the consistency of a running gait pattern, at least three gait cycles per trial with a double floating phase had to be observed. If there was no double floating phase during the running assessment, participants were classified as *walkers*.

Prior to the CGA, a standardized clinical examination was conducted including (a) muscle weakness, (b) spasticity, (c) selective motor control, and (d) passive RoM evaluation of the hip, knee, and ankle flexors and extensors. (a) Muscle weakness was measured by the Manual Muscle Testing (MMT) on a scale of 0 to 5 (Manikowska et al., 2018). (b) Muscle spasticity was measured using the Modified Ashworth Scale (MAS) on a scale of 0 to 4 (Bohannon and Smith, 1987). (c) Selective motor control was evaluated with the Selective Control Assessment of the Lower Extremity (SCALE) on a scale of 0 to 2 (Fowler et al., 2009). (d) Passive RoM was measured using a goniometer to the nearest 5 degrees (Viehweger et al., 2007).

Owing to the small sample size, results of muscle weakness, spasticity, selectivity, and passive RoM assessments were dichotomized as «normal» or «abnormal» based on the definition of each rating. Muscle strength rated at 5 or 4 on the MMT, a score of 0 on the MAS, and a selectivity score of 2 were considered as normal, as all those scores were described as no impairments (Bohannon and Smith, 1987; Fowler et al., 2009; Manikowska et al., 2018). The Thomas Test was ranked as normal ($<10^\circ$) or abnormal ($\geq 10^\circ$) based on reference values (Peeler and Anderson, 2008). For all other RoM assessments, values were categorized in 4 categories based on the 95% confidence interval (CI) of reference values (0: $\leq$ 5% CI, 1: between 5% and mean, 2: between mean and 95% CI, 3: $\geq$ 95% CI) (Soucie et al., 2011). A value between 1 and 3 was considered normal, and a value of 0 was abnormal.

At the end of the CGA, a functional evaluation comprising three tasks reflecting balance performance and muscle power was performed. For balance assessment, participants were asked to maintain a single-leg posture with their eyes open for at least 3-s. The task was evaluated on three levels; (a) unable to do the task on either leg, (b) able to do the task only on one side, and (c) able to do the task on either leg. The power tasks were a single-leg jump and a two-legged jump. The single-leg jump was evaluated on the same three levels as the balance task, i.e. (a) unable to do the task on either leg, (b) able to do the task only on one side, and (c) able to do the task on either leg, and the two-legged jump was evaluated as failure or success. The power tasks were rated as successful when the participant was able to lift off his/her feet from the ground. All functional assessments were evaluated on video recordings made during the CGA.

The most affected leg was kept for analysis. Before classifying into normal and abnormal values, individual impairment scores were grouped to joint specific impairment scores and subsequently grouped to a total composite score following the methodology of Papageorgiou et al. (2019). To ensure consistency across all impairments, the spasticity score was inverted (4 to 0, where a score of 4 means no spasticity). Per impairment, hip, knee and ankle joint scores were calculated as the sum of the agonist and antagonist impairment scores. In case of synergic muscles, a median score was calculated. A global score representing the sum of all scores per side was calculated. The lowest global score value was considered as the most affected leg.

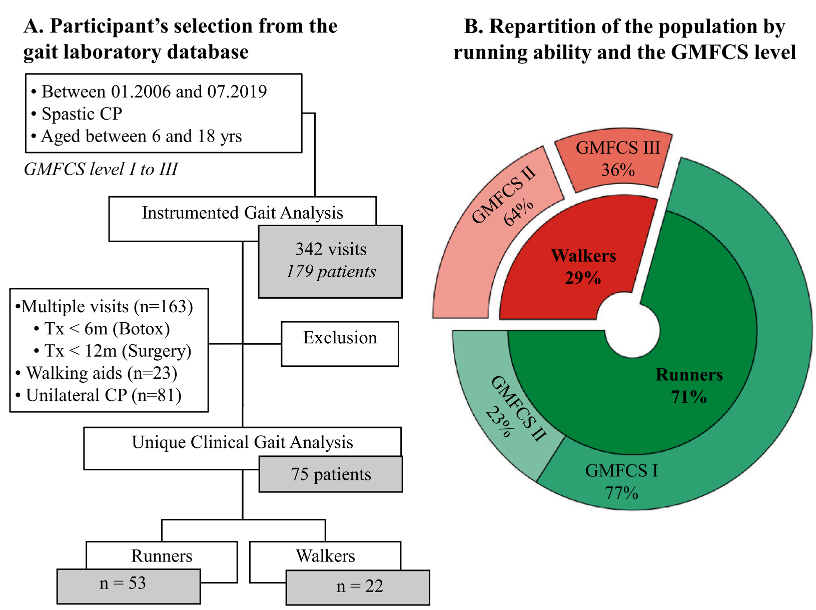
2.1. Statistical analysis

All participants were divided into a group of *runners* and *walkers*. Participants' characteristics, such as age, height, and mass were compared using an Unpaired Welch's *T*-Test. The BMI was categorized into four categories, i.e. underweight, normal weight, overweight and obese, following the reference of de Onis et al. (2007) and compared with a Chi-Squared Test between the groups. Clinical and functional characteristics were compared between the *runner* and *walker* groups by Chi-Squared tests with Yates continuity correction or the Fisher's Exact Test when the number of observations was insufficient (Kim, 2017). The unpaired group comparison was realized by a Mann-Whitney *U* Test, depending on the distribution of the data, for the global score and each joint and impairment score. The statistical significance level was $p < 0.05$. Statistical analyses were performed using R (version 3.6.2) and RStudio (version 1.2.5033).

3. Results

From the database, 342 visits were screened, corresponding to 179 patients. Based on the exclusion criteria, 75 children with a spastic BCP were included (Fig. 1A). On the 75 children, 53 participants (71%) were able to run and 22 participants (29%) did not achieve a double floating phase and therefore, were considered *walkers* (Fig. 1B). No differences were observed in the age, mass, height, and BMI between the *runner* and *walker* groups (Fig. 1C). A significant difference was observed in the GMFCS level and the sex between the two groups. Unsurprisingly, the *walker* group comprises children with a GMFCS level II (64%) and III (36%), and the *runner* group comprises children with a GMFCS level I (77%) and II (23%). A higher proportion of females was observed in the *walker* group (59%) compared to the *runner* group (30%).

The between-group comparison of joint and impairment scores is presented in Fig. 2. Significant differences were observed in all total joint scores with higher score values in the *runner* group. The total score for strength and selectivity was significantly higher for *runners*. The hip and knee spasticity scores were significantly higher in the *runner* group,



C. Clinical Characteristics

	Walkers (n=22)	Runners (n=53)	*p*
Sex (F)	13 (59%)	16 (30%)	**0.038***
Age (years)	11.1 [3.8]	10.6 [3.6]	0.191
Height (cm)	140.2 [17.7]	140.2 [16.2]	0.272
Mass (kg)	37.8 [16.3]	35.7 [16.3]	0.696
Body Mass Status			0.867
Underweight	3 (14%)	7 (13%)	1.000
Normal weight	13 (59%)	36 (68%)	0.642
Overweight	4 (18%)	7 (13%)	0.845
Obese	2 (9%)	3 (6%)	0.973
GMFCS			**0.000***
I	0 (0%)	41 (77%)	**0.000***
II	14 (64%)	12 (23%)	**0.002***
III	8 (36%)	0 (0%)	**0.000***

Data are presented as mean [SD] or n (%), F: Female, GMFCS: Gross Motor Function Classification System, Underweight (z-score < -2SD), Normal weight (-2SD ≤ z ≤ 1SD), Overweight (1SD < z ≤ 2SD), Obese (z > 2SD) from Onis et al, 2007.*: significant difference (p<0.05).

Fig. 1. Selection and characteristics of the population.

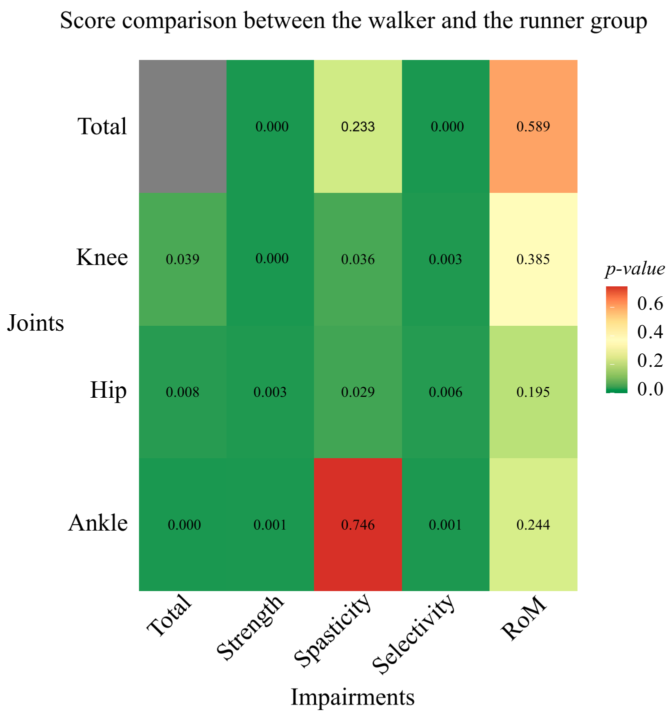


Fig. 2. Comparison of the joints and impairments clinical composite scores between the walker and runner groups.

but no differences were observed in the total spasticity and ankle spasticity scores. No differences were observed in the RoM score between the groups.

When comparing the clinical assessment classified as «normal» and «abnormal» between the two groups, significant differences in muscle strength, selectivity, and spasticity were observed (see Fig. 3 and supplementary data A). A higher proportion of «normal» values in muscle strength at the hip flexors (83% vs. 48%, $p = 0.006$), knee flexors (77% vs. 28%, $p = 0.001$), and ankle dorsiflexors (68% vs. 33%, $p = 0.014$) were observed in the *runner* group compared to the *walker* group. A higher proportion of «normal» muscle strength value was observed at the hip abductor in the *runner* group (63%) compared to the *walker* group (30%) ($p = 0.022$). The proportion of «normal» selectivity at the hip flexors (81% vs. 48%, $p = 0.011$), ankle plantiflexors (72% vs. 42%, $p = 0.043$), and dorsiflexors (67% vs. 23%, $p = 0.001$) was significantly higher in the *runner* group compared to the *walker* group. The proportion of «abnormal» spasticity at the knee extensors was significantly lower in the *runner* group (31%) compared to the *walker* group (59%) ($p = 0.045$). No differences were observed at the RoM for all joints tested.

All balance and power evaluations were significantly different between the groups (see Fig. 4 and supplementary file A). A large proportion of the *walker* group was unable to stand on one foot for at least three seconds on either leg (57% vs. 19%, $p = 0.013$). The majority of the *runner* group was able to stand on one foot on either leg (69%) compared to the *walker* group (14%) ($p = 0.001$). No participant in the *walker* group was able to perform the single-leg jump on either leg (0%) compared to the *runner* group (37%) ($p = 0.054$) and the majority of the *runner* group was able to perform a two-legged jump (94% vs. 45%, $p < 0.001$) compared to the *walker* group.

4. Discussion

The study aimed to compare biomechanical and neuromotor functions between a group of children with BCP who are able and unable to run. Our results show that children with BCP classified as *runners* were stronger and had greater selective motor control in flexor muscles than children classified as *walkers*. Moreover, children classified as *runners* had lower spasticity at the knee extensors, and globally, they perform better in balance and power assessment than children classified as *walkers*.

One of the main findings in the present study is the generalized differences in muscle strength and selectivity in flexor muscles between children who are able and unable to run. Previous studies suggested that overall muscle strength and selectivity were more important for walking performance than spasticity and RoM in this population (Desloovere et al., 2006; Ross and Engsberg, 2007). Our results are in line with these previous studies suggesting that muscle strength and selectivity are important parameters not only for walking but also for running capacity. Muscle strength and selectivity must be improved to develop running capacity in children and preserved through adolescence and adulthood as a decrease in gross motor function is reported in adults with CP (Hanna et al., 2009).

Our results show that hip and knee flexors strength, in addition to the ankle dorsiflexors, are determinants of running capacity in children and adolescents with BCP. In a typical running pattern, the forward momentum needed for running is produced by the swinging leg and arms rather than the stance leg like in a walking pattern (Dugan and Bhat, 2005). The action of the iliopsoas occurring during the initial swing phase to advance the limb forward would be crucial to realize the double floating phase required for running. Furthermore, during running, the hip joint is the principal power generator for forward propulsion in children with CP (Chappell et al., 2019b; Davids et al., 1998). Children with CP utilize a proximal strategy, i.e. hip flexors, to compensate for reduced plantarflexors power. Moreover, to increase speed from walking to running, children with CP increase their stride frequency rather than their stride length (Davids et al., 1998). The increased stride frequency in running is a particularity of children with CP as TD individuals increase their stride length firstly and increase their stride frequency at higher running speed (Chappell et al., 2019a). The strategy to increase stride frequency is achieved by the synergistic action of the iliopsoas, gluteus maximus, and hamstring of both legs that accelerate the leg in the swing phase (Dorn et al., 2012). Of all joints, the hip is the one where many differences were observed suggesting the importance of the hip joint for running ability in children with BCP.

Through the swing phase, adequate knee flexion and ankle dorsiflexion are needed to clear the foot as the limb advance forward. While walking, knee flexion during the swing phase can be restricted by the rectus femoris spasticity (Sutherland and Davids, 1993), which, based on our results and others (Böhm et al., 2018) is also related to running capacity in BCP. Even if the consequence of rectus femoris spasticity has not been specifically studied in running, the detrimental effect of rectus femoris spasticity on walking gait could also be present in running.

Our results also demonstrated that children who can run, have a greater selectivity at the ankle than children who are unable to run, meaning that the ability to fully extend the knee with the ankle in dorsiflexion or to flex the knee with the ankle in plantarflexion is a key determinant for running capacity. A previous study, reported that impaired selectivity is associated, while walking, with a reduction of step length, walking speed, knee flexion magnitude at initial contact, and a reduction of the knee angle at mid-stance in children with CP (Zhou et al., 2017). These previous results, as well as the observations reported in this study, suggest that an increased cadence, the use of hip flexors during the onset of the swing phase, and unrestricted knee flexion during the swing phase are critical parameters for achieving a running pattern in children with BCP.

Unsurprisingly, differences in power and balance tasks between youth who are able and unable to run were observed, as adequate muscle strength, selectivity, and RoM are required to perform a single leg jump or a one-leg standing. Moreover, in functional assessments such as the Gross Motor Function Measure (GMFM), running ability and the ability to jump are both considered as high motor functions in the fifth dimension: «Walking, Running and Jumping» (Russell et al., 2000).

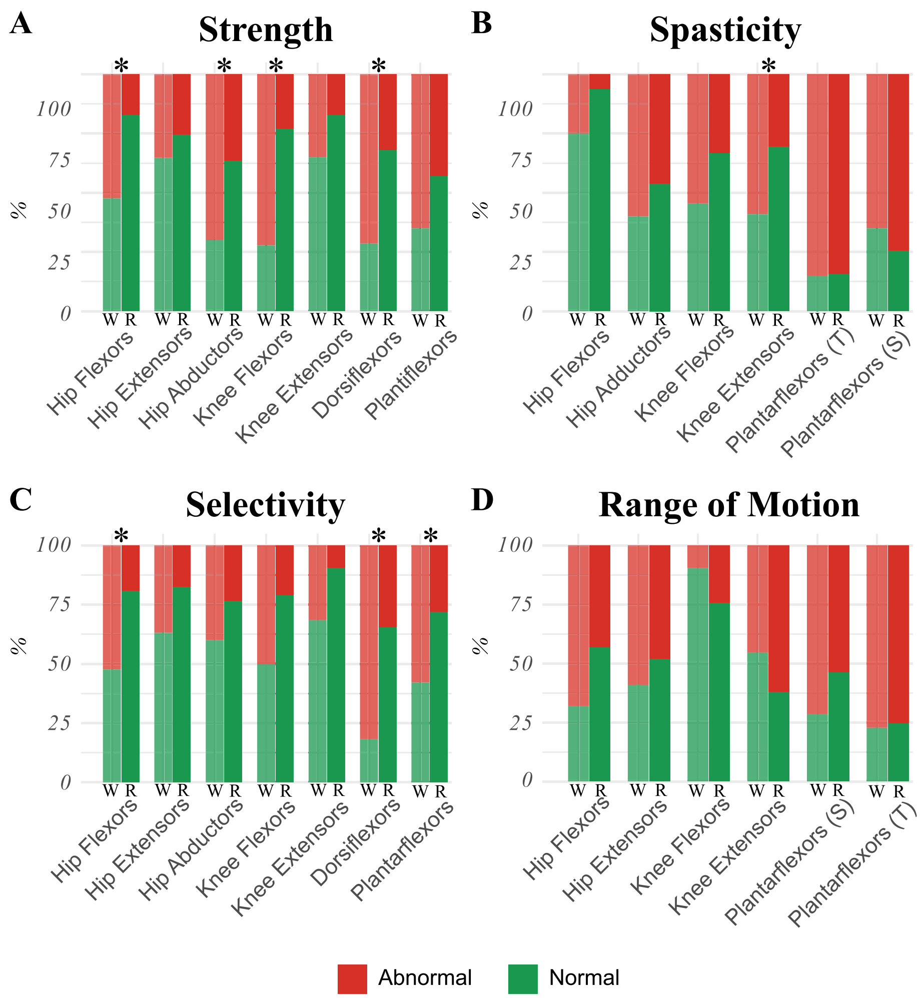


Fig. 3. Proportion of normal and abnormal values of muscle strength, selectivity, spasticity and range of motion between the walker and the runner group. The first column (lighter) represents the walker group (W) (n = 22) and the second column (darker) represents the runner group (R) (n = 53). * Significant difference (p < 0.05). (S): Soleus; (T): Triceps Surae.

Therefore, to jump or to stand on one foot seems as challenging as running.

Böhm et al. (2018), reported a detrimental effect of the rectus femoris spasticity with a reduction of chances of being able to run by 40% with each increase of one unit on the MAS (Böhm et al., 2018). They also reported a protective effect of gastrocnemius spasticity and no difference in muscle strength between children classified as *runners* and *walkers*. Despite the use of the MMT in both studies, our results suggest that muscle strength is a decisive parameter for running capacity. The studied population could partly explain this difference. Böhm et al., included children with unilateral and bilateral CP, with a majority of unilateral CP, and only children with a GMFCS level II. Despite the exclusion of unilateral impairments to specify the analysis of the present study, a more heterogeneous group of children (GMFCS level I-II-III) and the categorization of our variables as «normal» and «abnormal» could explain the different conclusions.

4.1. Study limitations

The results of the present study need to be interpreted with the current limitations in mind. The reliability and sensitivity of the clinical assessments can be discussed and their uses represent serious limits. Scores higher than 3 on the MMT are not considered objectives (Manikowska et al., 2018) and the MAS can only be valid when the increase in resistance to passive movement is exclusively associated with an increase in neural stretch reflex activity (Fleuren et al., 2010). Therefore Hand-Held Dynamometry and the Tardieu scale should be preferred. Further studies using more robust measurements should confirm the results of this study. In order to limit the low reliability of the clinical assessments, five experienced evaluators perform the clinical assessment since 2006. All the evaluators received a standardized training to lower the inter-rater variability. Clinical variables were categorized as «normal» / «abnormal» to limit the weaknesses reported on the clinical scales.

The functional tasks were part of a functional assessment. The psychometric properties of these evaluations are not known. Moreover, the functional assessment rated the task's capacity and not the performance. Evaluation rating performance (e.g. kinematics) could refine the results. A wider representation of the severity of impairments would allow to measure the performance and could help to specify the analysis. The selection of the first CGA visit corresponding to the inclusion and exclusion criteria could have lowered the average age of the studied population. Additional studies with older participants would possibly shed light or confirm clinical impairments influencing the ability to run. The mobility limitations increase with age, and it would give crucial elements to consider in therapeutic choices. Finally, a recruitment bias could be induced by the reason why children are recommended for a clinical gait analysis and therefore influence the representativeness of the study population (Pouliot-Laforte et al., 2022).

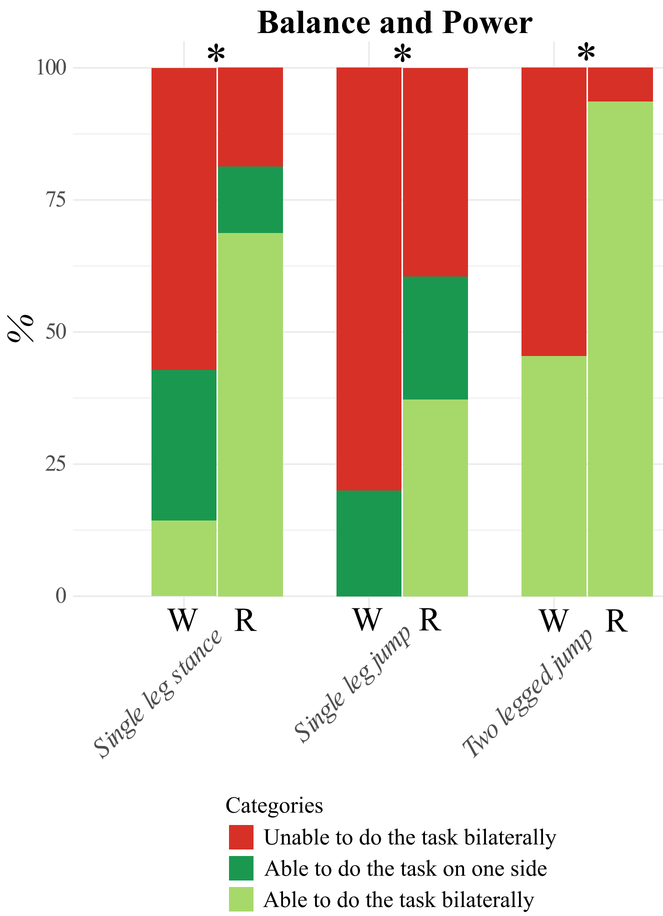


Fig. 4. Proportion of functional assessment results between the runner and walker groups. The first column represents the walker group (W) (n = 22) and the second column represents the runner group (R) (n = 53). * Significant difference ($p < 0.05$).

4.2. Conclusion

This study suggests that muscle strength and selectivity of the flexor muscles at the hip, knee, and ankle influence the capacity to run in youth with BCP, as well as spasticity at the rectus femoris. However, the results should be confirmed with more robust measurements. Passive RoM seems not associated with running capacity in this population. The results of the present study could help to better target interventions and increase the specificity of recommendations. Interventions should aim to maintain or improve muscle strength and adequate selectivity in flexor muscles at the hip, knee and ankle in order to maintain or improve high motor function in this population.

Funding

The work was supported by the Fonds de Recherche du Québec – Santé (FRQS). The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

CRediT authorship contribution statement

Annie Pouliot-Laforte: Conceptualization, Methodology, Writing – original draft, Writing – review & editing. **Anne Tabard-Fougère:** Conceptualization, Data curation, Methodology, Writing – review & editing. **Alice Bonnefoy-Mazure:** Conceptualization, Data curation, Methodology, Writing – review & editing. **Geraldo De Coulon:** Conceptualization, Writing – review & editing. **Stéphane Armand:** Conceptualization, Supervision, Writing – review & editing.

Declaration of Competing Interest

None.

Acknowledgments

The authors thank Antoine Poncet from the Clinical Epidemiology Service of the Geneva University Hospitals (Switzerland) for his statistical support.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.clinbiomech.2022.105817.

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