Archives of Rehabilitation Research and Clinical Translation (2020) 2, 100074

ACRM
AMERICAN CONGRESS OF REHABILITATION MEDICINE

Archives of Rehabilitation Research and Clinical Translation

Archives of Rehabilitation Research and Clinical Translation 2020;2:100074

Available online at www.sciencedirect.com

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Original research

Relation of Musculoskeletal Strength and Function to Postural Stability in Ambulatory Adults With Cerebral Palsy

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KEYWORDS
Bone mineral density;
Neurologic disorder;
Rehabilitation

Abstract *Objective:* To understand the relation of musculoskeletal strength and function to postural stability in ambulatory adults with cerebral palsy (CP) who have already developed muscle atrophy and osteoporosis.
Design: Two independent group comparison of adults with CP and those without it.
Setting: Laboratory study.
Participants: Thirteen adults with CP with sex (9 women: 4 men), age (21-62y), and Gross Motor Function Classification System I-III, and 13 sex-, age-, and body-weight-matched control participants completed our study (N=26).
Intervention: Not applicable.
Main Outcome Measure: Bone mineral density (BMD), structural or geometrical deformities (at the proximal region of the femur at the hip joint), and maximal muscular strength (forearm and thigh) were measured. The primary outcome measure was postural stability (balance measured using an automated balance system and a Berg Balance Test).
Results: Femoral BMD was significantly lower in the CP group compared to the control group, whereas BMD at lumbar and forearm regions was similar between groups. Geometrical angles, lengths, and diameters at the proximal femur were significantly lower in the CP group. There was a direct relation between BMD in the femoral neck and knee extension peak torque in the control group with no relation in the CP group. Although the control group did not show a relation between muscular strength and balance test, the CP group showed a significant linear relation among improving postural stability with greater levels of muscular strength.

List of abbreviations: BMD, bone mineral density; CP, cerebral palsy; DXA, dual-energy x-ray absorptiometry; GMFCS, Gross Motor Function Classification System; GT, greater trochanter.
Disclosures: none.
Supported by a part of Dr Jensen's Central Research, Scholarship and Creative Activity Grant and Undergraduate Research Grants by San José State University.
Presented to the American College of Sports Medicine, June 2, 2018, Minneapolis, MN; and Western Society for Kinesiology and Wellness, October 11, 2018, Reno, NV.
Cite this article as: Arch Rehabil Res Clin Transl. 2020;2:100074.

https://doi.org/10.1016/j.arrct.2020.100074

Conclusion: There were structural differences at the proximal femur and muscular weakness in adults with CP. In adults with CP, balance appears to be more influenced by structural alterations at the femur than muscular strength compared to the control group.

Cerebral palsy (CP) is the most common childhood motor disability and a neurodevelopmental disorder that is caused by lesions in specific areas of the brain.[1,2] CP is accompanied by abnormal musculoskeletal growth and development during childhood, which is also associated with impaired motor function and anomalous musculoskeletal structures as children get older.[3,4] Such motor impairments with abnormal muscular contracture, tone, and stiffness lead to functional limitations in activities such as walking and activities of daily living.[5] Even though neurologic insult is nonprogressive, adults with CP can experience worsening motor impairments because of aging effect on the skeletal muscles earlier than the general population.[5,6] In general, decrease in muscular strength, functional mobility, and dynamic postural stability are associate with a higher incidence of falls.[7,8] Therefore, muscular weakness driven from motor impairment in CP could expose adults with CP to more postural instability and fall-related injuries.

Low bone mineral density (BMD) is a significant clinical problem in individuals with CP, with a higher occurrence of fragility fractures compared to the general population.[9,10] Low BMD is significantly related to functional capacity and severity of physical disabilities (ie, Gross Motor Function Classification System [GMFCS]) in children with CP.[11,12] Because most of the studies have focused on children with CP, it is uncertain whether low BMD is related to the level of functional mobility and physicality in adults with CP. Trinh et al[13] reported that the locations of fractures in adults with CP are different than in children with CP.[13] Children with CP experience most of their fractures in the long bones of the lower limbs, whereas adults with CP are particularly vulnerable to hip fractures near the proximal region of the femur.[13-16] This indicates that differential pathophysiological alterations exist in adults with CP as they age. Thus, it is important to investigate whether skeletal strength in lower extremities has a differential effect on muscular strength and functional mobility in adults with CP.

Skeletal deformities (eg, joint contractures, scoliosis, hip displacement) get progressively worsen over time in CP, which likely causes further difficulties in physicality and mobility. Studies have found that the skeletal deformities in children with CP place them at an increased risk of low BMD and fractures.[17,18] Furthermore, joint contractures and deformities in the lower extremities may influence overall functional mobility related to balance.[19] However, it is still unclear whether the structural deformities in lower extremities affect the bone strength, muscular strength, and postural stability in adults with CP who have already progressed to muscle atrophy and osteoporosis.

Understanding the physiological interplay between the musculoskeletal system and balance in adults with CP is crucial because it may help them improve their functional mobility and physicality in a rehabilitation setting. Therefore, the purpose of this study was to investigate the relation of musculoskeletal strength to balance in adults with CP who have already progressed into muscle atrophy and osteoporosis. We hypothesized that skeletal deformities specific to the proximal femoral region at the hip joint are accompanied by low bone strength and low muscle strength in the lower extremities. Second, we hypothesized that musculoskeletal strength correlates with balance in adults with CP.

Methods

Participants

A total of 26 participants (13 adults diagnosed with CP along with 13 sex-, age-, body-weight-matched control participants) were recruited through CP centers in San José and in the Greater Bay Area. Both men and women of varying race and ethnic backgrounds were included. Individuals who were able to follow simple commands and walk with or without the use of an assistance apparatus (falling within classification levels I-III of the GMFCS) were included in the study. The rationale for such inclusion criteria was to make sure that participants could perform physical tests using their lower extremities. All experimental procedures and protocols conformed to the Declaration of Helsinki, and they were approved by the San José State University Institutional Review Board. Each participant and/or guardian signed a written informed consent prior to experimentation, and he or she completed a medical health history form. None of the participants had a history or symptoms of cardiovascular, renal, hepatic, or respiratory diseases and other neurologic disorders beside CP.

Experimental measurements/procedures

Anthropometric measurements including weight, height, and waist, and hip circumferences were obtained to characterize body size. Whole-body dual-energy x-ray absorptiometry (DXA)[a] was used to measure the mass and density of the bone, muscle, and fat. Regional DXA scans were performed to measure bone mineral content and BMD at the lumbar spine (from L1 to L4), the proximal femur, and the radial-ulnar regions. More specifically, bone mineral content and BMD at the ultradistal and the 33% of distance of the forearm regions on radius and ulnar bones were measured. All DXA scans were performed by the same, trained DXA technologist who is certified by the State of California.

Forearm muscular strength was assessed using a handgrip dynamometer over 3 trials of maximal voluntary isometric contraction. A Humac Norm Isokinetic Dynamometer system[b] was used to measure peak torque, work, and power during a full range of knee extension and flexion to assess muscular strength in hamstrings and quadriceps. Participants performed 2 sets of 3 repetitions of knee extension and flexion at 3 different velocities (ie, 90, 150, 210 deg/s) in a seated position with safety straps placed on the torso and the thigh. A 30-second rest interval followed each completed test for 2 sets of 3 repetitions.

Postural stability was assessed in 2 ways using a Biodex Balance System[c] and a Berg Balance Test. A modified protocol using the limits of stability test from the Biodex Balance System was performed in an upright position with the back unsupported. The best scores of the 3 separate tests and time to complete the test were reported following all directional shifts (eg, forward, backward, left, right, forward/left, forward/right, backward/left, backward/right) in trunk movement. The Berg Balance Test measures balance ability among older adults and is a reliable method to test postural stability. Following their well-established criteria, participants performed various functional activities such as reaching, bending, transferring, and standing. Each item was scored along a 5-point scale from 0 to 4.

Skeletal structure was assessed geometrically using a customized function during BMD analysis on DXA software. Skeletal deformities at the proximal femur were identified by geometrically measuring angles from a centerline through the femoral neck and head (1) to the proximal greater trochanter (GT), (2) to distal GT, and (3) to the lesser trochanter (supplemental fig S1, available online only at http://www.archives-pmr.org/). In addition, lengths and diameters of the femoral neck and head, and shaft diameter were identified.

Statistical analysis

The means of peak torque, work and power, and the best score of handgrip strength were averaged. All data are presented as mean ± standard error of estimate. Comparisons between the CP versus control groups on all variables were made using independent sample unpaired *t* tests. Pearson product-moment correlation coefficients were calculated to indicate the strength of relations for geometrical measures of skeletal geometry, BMD, and forearm and leg muscular strength, and postural stability. Statistical analyses were conducted using IBM SPSS Statistics version 24.[d] Statistical significance was set at $P<.05$.

Results

Anthropometric differences

Control and CP groups had similar characteristics except for height. Even though waist and hip circumference measures were not significantly different between groups, waist-to-hip ratio was greater in the CP group than in the control group (table 1). All CP participants had spastic CP. Two participants had mixed types with spastic and dyskinetic CP. All participants with CP were classified in GMFCS I-III (see table 1).

Musculoskeletal structure, strength, and postural stability

The CP group had significantly smaller angles to the proximal GT and the distal GT than the control group (fig 1A). There were no significant differences in the angles to the lesser trochanter between groups (see fig 1A). Although the diameter of the femoral neck and femoral head was similar in both groups, the length of the femoral neck and the diameter of the shaft were shorter in the CP group than in the control group (figs 1C-E).

Skeletal strength at the femur was significantly lower in the CP group than in the control group ($P<.05$) (table 2), whereas the markers at the lumbar spine (L1-L4) and forearm regions (both the radius and the ulna) were not significantly different between groups (see table 2). After the World Health Organization criteria for defining osteoporosis (BMD that lies 2.5 standard deviations or more below the average value for young healthy individuals with an age of 30y) at any regional sites, 10 CP and 3 control participants had osteoporosis.

The CP group demonstrated significant weakness across all measures of muscular strength for both the forearm handgrip and the knee extensors and flexors at 90, 150, and 210 deg/s (table 3). Peak torque across all angular velocities was significantly lower in the CP group than in the control group. Peak torque during knee extension at 90 deg/s was significantly higher than the torque at 150 or 210 deg/s regardless of the group. However, peak torque, work, and power during knee flexion remained constant for all angular velocities in the CP group, whereas the control group showed dose-response relations for different angular velocities (see table 3). Handgrip strength was significantly lower in the CP group than in the control group (see table 3).

Table 1 Participant characteristics

Characteristics	Control Group	CP Group
Age (y)	34.3±4.5	37.3±4.2
Sex (men/women)	4/9	4/9
Weight (kg)	66.2±3.4	65.2±5.5
Height (cm)	169.6±2.8	157.3±3.2*
BMI (kg/m^2)	22.9±0.8	26.6±2.5
Waist circumference (cm)	78.8±2.9	86.1±4.9
Hip circumference (cm)	97.8±2.2	97.4±4.2
Waist-to-hip ratio	0.80±0.1	0.88±0.1*
Types (spastic/mixed)	-	11/2
GMFCS level (I/II/III)	-	2/7/4
Unilateral/bilateral	-	2/11
Use of botulinum toxin	-	1

NOTE. Values are mean ± SE.
Abbreviation: BMI, body mass index.
* $P<.05$ vs control group.

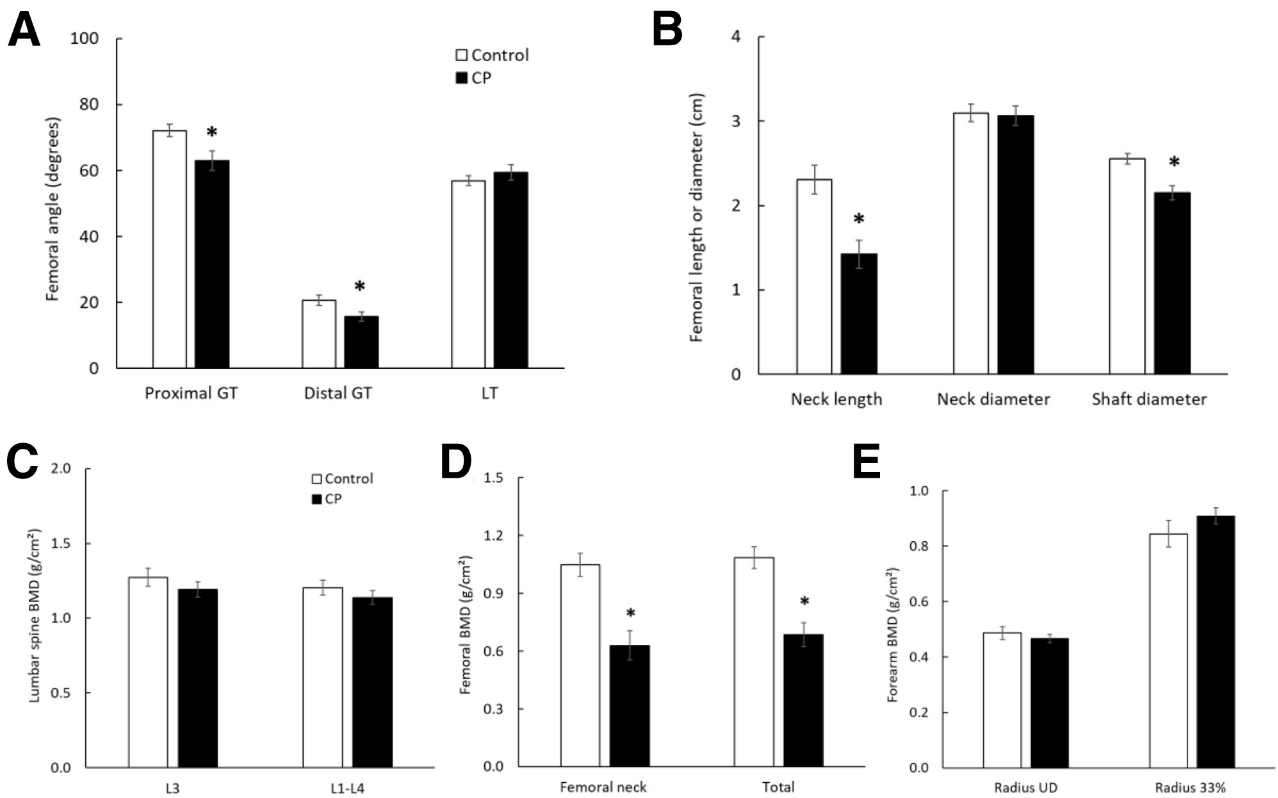

Fig 1 Summary of data showing the group differences in skeletal structure of the proximal femur at the hip joint (angles from the proximal GT, the distal GT, LT retrospect to the centerline of the femoral neck [A], and femoral neck length, neck diameter, shaft diameter [B]), and BMD in various regions (lumber spines [C], proximal femur [D], forearm [E]). **P*<.05 versus control group. Abbreviation: LP, lesser trochanter.

Overall postural stability was significantly lower in the CP group compared to the control group. For all balance scores after the limit of stability test, the CP group had significantly lower scores in all directions, and it took longer to complete the limits of stability test than the control group (table 4). Berg balance score was significantly

Table 2 Characteristics of skeletal strength in lumbar spine, proximal femur, and forearm in participants with and without CP

Characteristics	Control Group				CP Group			
	BMC (g)	BMD (g/cm^2)	T Score	z Score	BMC (g)	BMD (g/cm^2)	T Score	z Score
Lumbar spine								
L_1	13.4±0.9	1.06±0.04	−0.72±0.32	−0.47±0.25	11.8±0.7	1.06±0.05	−0.78±0.44	−0.82±0.39
L_2	15.5±1.1	1.16±0.05	−0.44±0.39	−0.17±0.32	12.7±0.6*	1.11±0.05	−0.92±0.45	−0.95±0.42
L_3	18.4±1.4	1.27±0.06	0.41±0.44	0.67±0.38	15.4±0.8	1.19±0.05	−0.32±0.42	−0.33±0.42
L_4	20.7±1.5	1.28±0.06	0.35±0.42	0.61±0.38	16.0±1.2*	1.19±0.04	−0.35±0.35	−0.38±0.31
L_1-L_4	68.8±4.5	1.20±0.05	−0.02±0.38	0.25±0.32	56.1±3.1*	1.14±0.05	−0.57±0.39	−0.60±0.35
Left femur								
Neck	5.0±0.5	1.05±0.06	0.05±0.41	0.32±0.39	3.08±0.46*	0.63±0.08*	−2.89±0.55*	−2.92±0.53*
Total	35.1±2.8	1.08±0.06	0.31±0.36	0.54±0.34	19.4±2.2*	0.68±0.06*	−2.71±0.45*	−2.85±0.47*
Forearm								
Radius UD	1.92±0.12	0.49±0.02	0.11±0.41	0.29±0.39	1.81±0.09	0.47±0.01	−0.19±0.37	−0.32±0.35
Radius 33%	2.31±0.14	0.84±0.05	−0.38±0.26	−0.19±0.24	2.28±0.13	0.91±0.03	0.08±0.37	−0.07±0.31
Radius total	11.08±0.81	0.69±0.03	−0.22±0.35	−0.04±0.32	9.67±0.66	0.68±0.02	−0.18±0.41	−0.33±0.36
Ulna UD	0.84±0.08	0.36±0.02	-	-	0.72±0.05	0.36±0.02	-	-
Ulna 33%	2.10±0.14	0.86±0.04	-	-	1.98±0.12	0.93±0.04	-	-
Ulna total	7.50±0.59	0.66±0.03	-	-	6.26±0.43	0.68±0.03	-	-

NOTE. Values are mean ± SEM.
Abbreviations: BMC, bone mineral content; UD, ultra distal.
* *P*<.05 vs control group.

Table 3 Summary of muscular strength of forearm during isometric maximal handgrip contraction and of thigh during knee extension and flexion in adults with and without CP

Characteristics	Control Group			CP Group		
Handgrip strength (kg)	34.0±2.7			17.5±4.8*		
Leg strength	Peak torque (ft-lb)	Work (ft-lb)	Power (W)	Peak torque (ft-lb)	Work (ft-lb)	Power (W)
Knee extension						
90°/s	85.7±12.2	101.8±14.5	110.9±17.2	29.0±8.2*	32.7±12.2*	44.5±18.0*
150°/s	69.7±11.2†	83.5±13.0†	120.4±24.4†	20.7±6.4*,†	24.0±9.8*,†	45.3±21.4*
210°/s	60.0±8.9†	70.5±10.2†	157.1±24.6†	15.7±5.0*,†	16.8±7.8*,†	36.3±16.9*,†
Knee flexion						
90°/s	55.2±6.8	70.7±8.8	70.9±11.2	13.0±3.9*	16.0±7.1*	23.0±12.0*
150°/s	48.6±6.7†	55.9±8.9†	102.4±14.7†	11.5±3.6*	12.9±6.4*	24.1±13.1*
210°/s	44.2±5.5†	52.4±6.5†	116.2±15.7†	10.2±3.0*	10.4±5.2*	21.8±11.5*

NOTE. Values are value ± SEM.
* $P<.05$ vs control group.
† $P<.05$ vs 90 deg/s.

lower in the CP group compared to the control group (21.9±6.0 CP vs 56.0±0.0 control) ($P<.05$).

The relations among skeletal structure and BMD, muscular strength, and balance

There was a weak but direct relation between BMD at 33% of the radius in the forearm and maximal handgrip isometric strength in the CP group, with a direct relation in the control group (fig 2A). Meanwhile, there was a strong linear relation between BMD in the femoral neck and knee extension peak torque at 90 deg/s in the control group, with no observed relation in the CP group (fig 2B). Although the control group did not show a relation between muscular strength and balance, the CP group showed a significant linear relation indicating greater muscular strength with better balance (fig 3A). Neither group showed any relation between BMD in the femoral neck and balance (fig 3B). There was an inverse relation between skeletal structure (eg, angles to proximal GT) and balance in the CP group (fig 3C). Of note, a lower amount of time to complete the test correlated to better stability and balance.

Table 4 Summary of postural stability for directional movement during limit of stability test

Characteristics	Control	CP
Forward	70.3±5.4	40.5±6.7*
Forward goal (%)	101.8±8.3	61.9±9.3*
Backward	76.2±3.5	40.1±9.2*
Backward goal (%)	214.6±25.3	119.4±31.1*
Left	72.3±3.8	33.8±7.0*
Left goal (%)	106.2±7.5	52.3±9.7*
Right	67.5±3.4	39.5±7.0*
Right goal (%)	94.7±6.0	60.3±9.4*
Overall score	59.1±3.2	28.8±6.1*
Overall goal (%)	88.1±5.5	46.2±8.5*
Time to complete (s)	29.9±1.7	81.3±17.7*

NOTE. Values are mean ± SEM.
* $P<.05$ vs control group.

Discussion

The purpose of this study was to investigate alterations in musculoskeletal strength and function, and their relation to balance in adults with CP. We conclude that adults with CP demonstrated structural alterations at the proximal femur which influence balance differently compared to the control group. Our evidence for the main findings is as follows: (1) adults with CP have skeletal deformities at the proximal femur and lower BMD in the femur than the control group, with similar BMD in lumbar spine and forearm regions; (2) femoral BMD may not significantly influence muscular strength in adults with CP; instead, structural deformities appeared to influence muscular strength in adults with CP; (3) both deformities and muscular strength affect balance in only the CP group but not in the control group.

Although CP is a lifelong condition, most research has focused on the pediatric issue; very little research has investigated the pathophysiological aspects of adults with CP. In addition, even though the worsening of structural deformities on various joints and bones has been identified in adults with CP, the relations to muscular strength and functional mobility have not been acknowledged. In the current study, we attempted to quantify structural deformities by measuring geometrical characteristics at the proximal femur, where the main osteoporotic process occurs in adults with CP. We also identified differential effects of deformity on postural stability in the CP population compared to the control group. Noble et al[20] found that the mechanical properties of the femur were associated with reduced thigh muscle volume in adolescents with CP. Overall, they found that bone strength was related to BMD; however, the skeletal strength was determined predominantly by bone geometry in the CP population.[20] Other studies suggest that the ability of a bone to resist forces is inversely proportional to bone length and directly related to bone diameter.[21] The reduced bone strength and

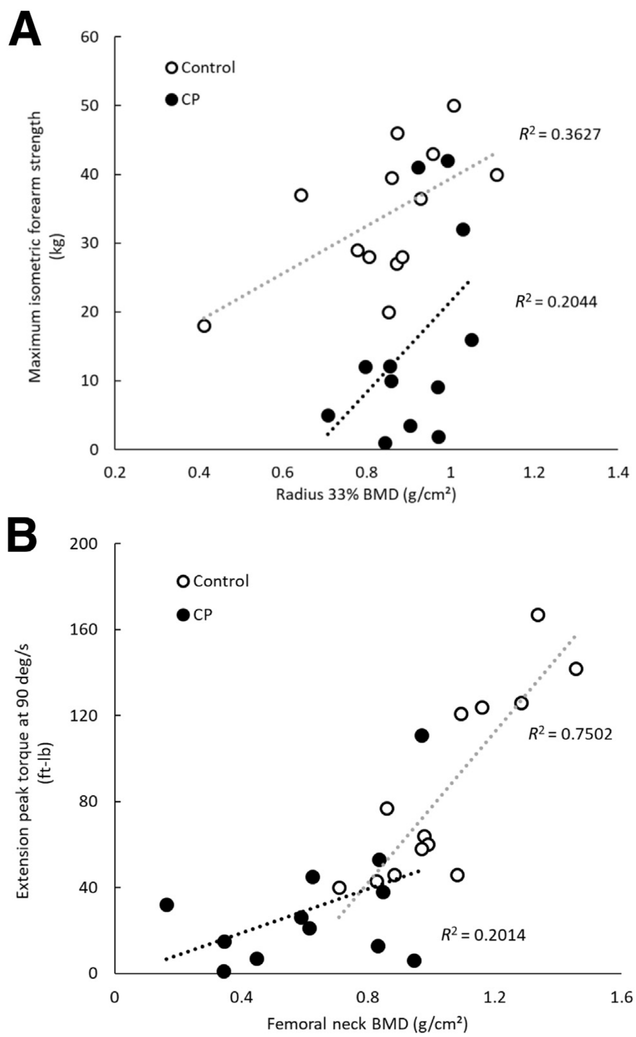

Fig 2 The relation between BMD at 33% region of the radius and maximal isometric handgrip strength (A), and BMD in the femoral neck and knee extension peak torque at 90 deg/s (B) in control and CP groups.

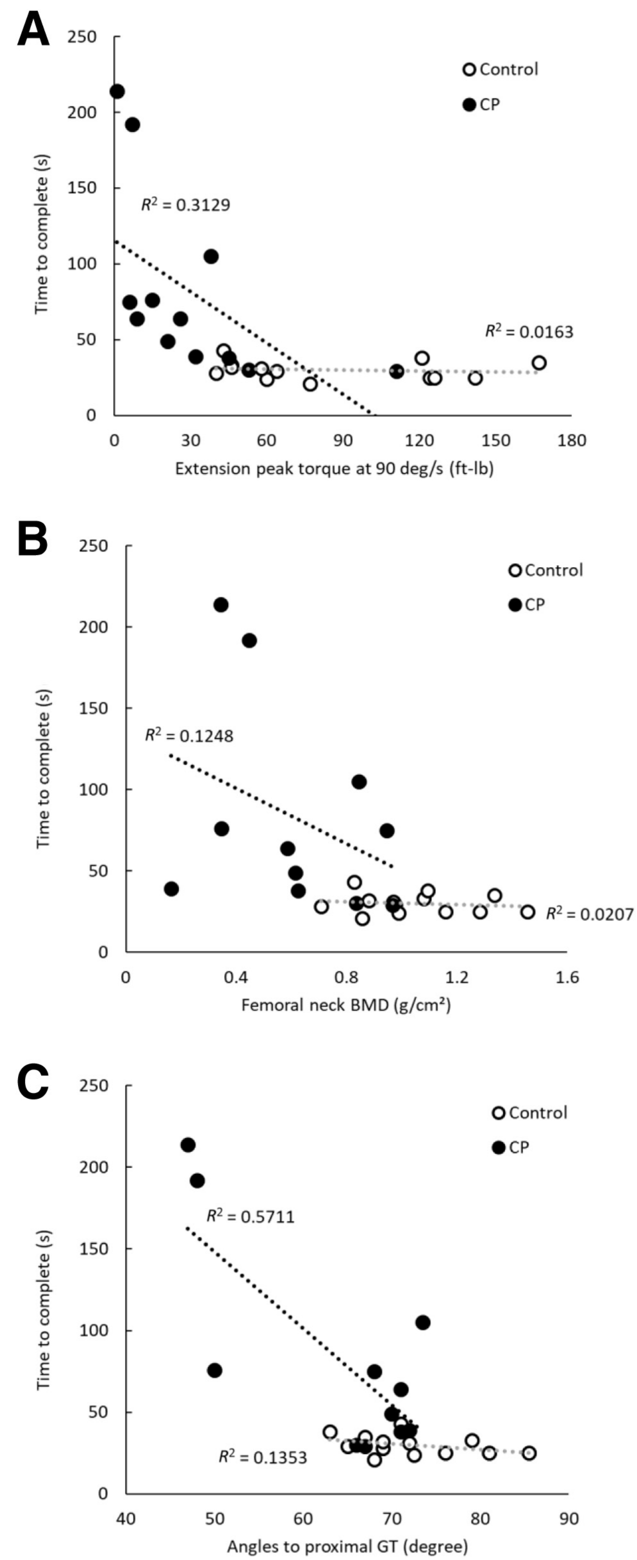

Fig 3 The relation between the total time taken to complete the limits of stability balance test and knee extension peak torque at 90 deg/s (A), BMD at the femoral neck (B), and angles to proximal GT at the femur (C).

increased fracture rate in adults with CP could be due to the attenuated structural property of the bone.[20-22] Our data support the importance of skeletal deformities at the proximal femur, which may influence muscular strength and balance in adults with CP. Thus, discovering ways to improve skeletal deformities in the CP population may result in improving functional mobility throughout the lifetime in adults with CP.

Our data showed that BMD in adults with CP was not related to muscular strength or balance, whereas those with higher BMD exhibited higher muscular strength in the control group. We observed that participants within the CP group who had higher BMD appeared to have higher muscular strength. This level was similar to participants within the control group who had lower muscular strength with similar BMD levels. We speculated that a certain threshold of BMD may be necessary to show a linear relation between BMD and muscular strength. Unpublished data from our laboratory support that adults with CP who improved BMD after resistance training gained muscular strength significantly; a linear relation between BMD and muscular strength began to appear in CP group (Abstract 8007, ACSM 2019). There is a possibility that improvement of muscular function and BMD with exercise training in adults with CP may directly improve postural stability, which could eventually increase functional mobility. Indeed, Gillett et al[23] documented the importance of resistance or strength training on the musculoskeletal system, in that muscular function may correlate with musculoskeletal morphology and architecture in individuals with CP. Consequently, enhancement of the musculoskeletal system seems to be a promising approach to harness BMD and improve functional mobility in CP population.

Previous studies in children with CP have reported equivocal results because some studies found a correlation between muscular strength and their level of GMFCS, whereas other studies on young adults with CP indicated that greater strength did not result in an improvement of mobility (ie, walking).[24-26] Our study suggests that adults with CP who have better postural stability and functional mobility are due to their greater leg muscular strength, whereas healthy adults showed similar stability no matter how strong their leg strengths are. We believe that this result has promise for further improvement of functional mobility in adults with CP. Further research on the effect of well-designed resistance training is necessary among adults with CP to determine whether such interventions not only improve their muscular function but also improve overall functional mobility.

Rose et al[27] reported abnormal variations in the size of muscle fibers and altered distributions of Type I and II fibers in hamstrings and quadriceps in lower extremities in children with CP. Although we did not measure direct muscular fiber types in our study, we identified differential levels of leg muscular strength between knee extensors (eg, quadriceps) and flexors (eg, hamstrings) in that knee flexors produced similar torque, work, and power regardless of the speed. We speculate that knee extensors and flexors may have differential stages in muscular atrophy and/or muscle pathology in adults with CP. Hamstrings are important for initiating a gait cycle.[28] Smith et al[29] found that hamstring contractures in children with CP result from a stiffer extracellular matrix and increased sarcomere length. Our data showed that strength of flexor muscles (eg, low peak torque and power) was similarly weak in the CP group, suggesting that muscular function on hamstrings among adults with CP may have already reached the weakest point which may have caused their lower postural stability. Thus, the weakening of specific muscles and skeletal alterations could lead to additive effect on functional mobility in adults with CP. Further studies on this subject may necessary to determine the characteristics of CP about musculoskeletal pathology and functional capacity.

Genu recurvatum is common in individuals with CP, which causes overall changes in walking and movement patterns.[30] Severe recurvatum may occur in patients with weak hamstrings.[30] Skeletal structure in lower extremities is critical for maintaining and producing proper body movement. Our study participants have varying degrees of contractures, scoliosis, and hip displacement. These structural factors are all important in maintaining good posture, balance, and functional mobility from a biomechanical standpoint. We attempted to determine how structural alterations at the proximal femur relate to muscular strength in the lower extremities and postural stability in adults with CP. Future investigations about the differential roles of the distal femur and overall movement patterns in the conjunction of hip and knee joints for proper movement may help inform exercise training regimens and introduce nonpharmacologic treatment for individuals with CP.

Study limitations

Findings should be interpreted in the context of study limitations. We are aware of the small sample size, with 13 participants in each group. The size of the study depends on the magnitude of the expected effect size when comparing characteristics between $\geq$2 groups of participants.[31] Because our study showed very significant group differences in the main outcomes, we felt safe to report our results despite our small sample size per group. However, increasing the sample size may help show better correlations of individual variables in our conclusion.

We did not identify structural abnormality of muscles at the tissue level. A few studies suggest that muscle volume and fascicle lengths tend to be reduced in children with CP compared to typically developing peers, whereas sarcomere and tendon lengths are longer in children with CP.[23,27,29] However, there is limited information available if this maintains true for physiologically mature adults with CP. Future studies might investigate additional architectural alterations of the musculature and tendons at the tissue level in adult CP populations.

This study has potential limitations on reporting various structural alterations existed in participants with CP. Even though we identified other skeletal abnormalities, such as joint contractures, scoliosis, and hip displacement, in participants with CP, we did not include all individual differences in our result section. Our study only reported structural deformities on the proximal region of the femur with geometrical measures because we focused on the muscular strength in the lower extremities and its relation to postural stability. Investigating the relations of additional structural deformities, other than at the femur, and muscular strength with postural stability could strengthen our future studies.

Last, our study did not fully account for all medications that may have influenced outcomes. Anecdotal and limited studies have reported that use of antispastic drugs (eg, botulinum toxin) can decrease muscular function, but may improve functional mobility.[32] There was only 1 participant out of 13 in the CP group who reported the use of Botox. Most of the participants in our study used other medications, such as anticonvulsant, antihypertensive, and antidepressant drugs. In addition, some of the participants with CP take daily calcium and vitamin supplements. We cannot rule out the effect of calcium supplements related to the level of BMD and whether it has additional effects on muscular strength and functional mobility.

Conclusions

Our findings suggest that skeletal deformities at the proximal femur and leg muscular strength appear to influence balance in adults with CP, whereas no relation was detected in the control group. In addition, BMD may not influence leg muscular strength in CP, whereas there is a strong linear relation in the control group. Thus, we conclude that there are pathophysiological and anatomical differences in adults with CP, and their balance can be differently influenced by structural alterations at the lower extremities compared to the control group.

Suppliers

a. Dual-energy x-ray absorptiometry; Lunar iDXA, GE.
b. Humac Norm Isokinetic Dynamometer; Computer Sports Medicine Inc.
c. Biodex Balance System; Biodex Medical Systems, Inc.
d. SPSS statistical software; IBM.

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