

Promotion of Physical Fitness and Prevention of Secondary Conditions for Children With Cerebral Palsy: Section on Pediatrics Research Summit Proceedings

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Inadequate physical fitness is a major problem affecting the function and health of children with cerebral palsy (CP). Lack of optimal physical activity may contribute to the development of secondary conditions associated with CP such as chronic pain, fatigue, and osteoporosis. The purpose of this article is to highlight the content and recommendations of a Pediatrics Research Summit developed to foster collaborative research in this area. Two components of physical fitness—muscle strength and cardiorespiratory fitness—were emphasized. Although there is evidence to support the use of physical fitness interventions, there are many gaps in our current knowledge. Additional research of higher quality and rigor is needed in order to make definitive recommendations regarding the mode, intensity, frequency, and duration of exercise. Outcome measurements have focused on the body functions and structures level of the *International Classification of Functioning, Disability and Health* (ICF), and much less is known about effects at the activities and participation levels. Additionally, the influence of nutritional and growth factors on physical fitness has not been studied in this population, in which poor growth and skeletal fragility have been identified as serious health issues. Current intervention protocols and outcome measurements were critically evaluated, and recommendations were made for future research.

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Section on Pediatrics Research Summit Participants and Section on Pediatrics Research Committee Task Force (see Appendixes for participants and member sites and investigators on pages 1513–1514).

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In 2003, the American Physical Therapy Association (APTA) Section on Pediatrics and its Research Committee determined that there was a critical need to identify and promote effective physical fitness interventions for children with cerebral palsy (CP). Thus, a research summit was initiated by the Research Committee, funded by the Section on Pediatrics, and implemented in 2004 with the purpose of fostering research in the area of physical fitness in children with CP. The specific aims of the Research Summit were: (1) to assimilate current and emerging knowledge on the physical fitness of children with CP and secondary complications associated with inactivity, (2) to identify gaps in research, (3) to define and refine types of intervention and outcome measures to be used in research, and (4) to discuss ways to foster and develop innovative multi-site research programs. Research Summit participants (Appendix 1; see Appendix 2 for the Section on Pediatrics Research Committee Task Force) were selected through a competitive process emphasizing relevant research. Invited participants included consumers, representatives from funding agencies, and experts in the areas of physical therapy, adapted physical education, biomechanics, early childhood development, exercise physiology, neurology, nutrition and growth, occupational therapy, orthopedics, and psychology.

Existing evidence and recommendations were discussed using the *International Classification of Functioning, Disability and Health* (ICF) framework,^{1,2} a classification containing 3 domains of human function: body functions and structures, activities, and participation. *Body functions* refers to the physiological function of body systems, whereas *body structures* refers to anatomical parts of the body. Assessment of a child's physical fitness at this level would include assessment of muscle strength (force-generating capacity)

and oxygen consumption ($\dot{V}O_2$). *Activities* refers to the performance of a task or action by the whole person, such as standing, walking, running, and jumping. *Participation* refers to a child's involvement in life situations, including recreation and sport activities. Two contextual factors also are included in this framework: environmental and personal. Environmental factors are the physical, social, and attitudinal environment in which a child lives, whereas personal factors include the background of a child's life and psychological factors. This report focuses on critical evaluation of interventions and outcome measurements in 2 areas of physical fitness: muscle strengthening and cardiorespiratory fitness. A review of growth, nutrition, and secondary conditions specific to CP is included, as these factors influence the design of safe and appropriate physical fitness programs.

Need for Research

Children with CP are weaker,^{3–8} have less endurance,^{9–13} and exhibit reduced physical activity levels^{14–16} compared with children without CP. These findings are of concern because the benefits of physical activity and exercise to overall health are well known and people with disabilities are less likely to engage in physically healthy lifestyles compared with people without disabilities.¹⁷ Additionally, inactive adults with disabilities exhibit increased severity of disease and reduced overall health and well-being.¹⁸ Impairments such as weakness,¹⁹ muscle spasticity,²⁰ and deficient balance²¹ make it difficult for children with CP to participate in sport and play activities at a level of intensity sufficient to develop and maintain normal physical fitness levels. Research is needed to identify safe and effective methods to improve physical fitness in this population.

Muscle Strength Importance of Muscle Strength in Children With CP

Muscle weakness is a primary impairment in children with CP, as the diagnosis is dependent upon injury to a region of the brain responsible for movement. Research has shown that muscle force production can be improved in children with CP^{19,22-31} and that improved strength can translate into functional gains.^{23,26,28,30,32,33} The principles used for strength training, in terms of weight progression and specificity of training, are similar to those for people without disabilities.³⁴ Preliminary information on safety and lack of adverse effects supports strength training in children with disabilities, including CP.^{35,36} In particular, the concern that the performance of strengthening exercises will increase spasticity³⁷ appears to be unfounded.^{25,28,30,38,39}

Evidence of Muscle Weakness in Children With CP

Muscle weakness associated with the spastic form of CP is the aspect of physical fitness that has been studied to the greatest extent. Insufficient force generation has been attributed to decreased central activation or neuronal drive,^{4,6-8} inappropriate coactivation of antagonist muscle groups,^{4,7,8} secondary myopathy,⁴⁰⁻⁴² and altered muscle physiology.⁸ The lower-extremity musculature, in particular, has been the focus of many of these studies. Greater weakness has been reported in the distal musculature, as compared with the proximal musculature.^{3,4,7,8,43}

Wiley and Damiano⁷ found reduced hip, knee, and ankle muscle strength bilaterally in children with spastic diplegic CP and on the hemiplegic side in children with spastic hemiplegic CP as compared with a control group of children without disabilities. Weakness also was documented in the nonhemiplegic lower extremity. Similarly, Stackhouse et al⁸ found that

children with spastic diplegic CP produced 56% and 73% less knee extensor and ankle plantar-flexor force, respectively, compared with participants without disabilities.

During dynamic contractions, similar strength deficits have been reported for the ankle and knee musculature.⁴⁴ Overall, greater strength impairments were found for concentric versus eccentric contractions and faster versus slower speeds of movement.⁴⁴ Elder et al⁴ found reduced plantar-flexor and dorsiflexor muscle cross-sectional areas in children with CP when compared with an age- and weight-matched control group. Weakness was attributed to decreased neuronal drive because the children with CP were unable to produce joint torque levels commensurate with muscle cross-sectional areas.⁴ Using surface electromyography, Stackhouse et al⁸ demonstrated activation deficits of 39% for the quadriceps femoris muscles and 49% for the triceps surae muscles in children with spastic diplegic CP compared with age-matched participants without disabilities.

Studies^{4,7,8} have shown that coactivation of antagonist muscle groups is excessive in children with CP, contributing to reduced net joint torque production. For example, during isometric activation of the quadriceps femoris muscles, the ratio of quadriceps femoris to antagonist semitendinosus muscle activity was 0.73 for children with CP versus 0.22 for children who were developing typically.⁸ In addition to altered neural activation, secondary myopathy⁴⁰⁻⁴² and altered muscle physiology (differing muscle force-frequency relationship and fatigue properties)⁸ have been identified in spastic muscle and may contribute to muscle weakness.

Muscle Strengthening Interventions

Muscle strength training is the area of physical fitness that has received the most attention. Significant gains ($P < .05$) in muscle strength using a progressive resistance exercise approach have been documented,^{19,24-31} and effect sizes for strength changes ranging from 1.16 to 5.27 have been reported.⁴⁵ Using Cohen's criteria, effect sizes greater than .80 are considered to be "large."⁴⁶ At the activities level, improvements were reported for the Gross Motor Function Measure (GMFM),^{23,28,30} self-selected walking speed,^{23,33} walking cadence,^{23,30,33} and the Timed "Up & Go" Test.³² Children reported that they felt better about their appearance following a community-based strengthening intervention.²⁴ In contrast, Dodd et al⁴⁷ reported that self-concept, as measured by the scholastic competence section of the Self-perception Profile for Children, decreased in the exercise group, when compared with a control (no exercise) group, following a home exercise program. Possible explanations included differences between home and community programs, time away from homework during home exercise, and the inclusion of younger children in the latter study.⁴⁷

Strengthening exercise protocols varied considerably across studies. Muscle groups were targeted using free weights,^{19,22-24,27} strength training machines,^{24,30,31} and isokinetic exercise.^{28,29} Dodd et al²⁵ instructed children in 3 exercises targeting specific lower-extremity muscle groups and added progressive resistance via a backpack with weights. Thorpe et al³² provided progressive resistance to lower-extremity muscle groups using variable resistance of moving in water at different speeds as well as using fins, Hydro-Tone boots,* and ankle weights

* Hydro-Tone Fitness Systems Inc, 22895-E Savi Ranch Pkwy, Suite E, Yorba Linda, CA 92887.

during aquatic exercise. Frequency for these programs was typically 3 times per week for a duration of four,²⁶ six,^{19,23,25,29,30} eight,^{27,28,31} or 10^{24,32} weeks.

Although these studies provide convincing evidence that strengthening exercises are effective for people with CP, additional research is needed. A 2002 review of the effectiveness of muscle strengthening programs for people with CP revealed that only 10 studies in this area had sufficient methodological quality for further analysis.⁴⁵ Only one study²⁹ was a randomized controlled trial. Eight of these studies concluded that resistive exercise improved strength in this population.^{19,23,24,27-29,31,48} The total number of subjects across all studies was 126, with a range in age from 3 to 30 years. Only 3 studies addressed activity-related outcomes such as gait speed,^{23,28} GMFM scores,^{23,28} and wheelchair propulsion.³¹ Only one study²⁴ examined self-perception, and none assessed participation.

The magnitude of the effect varied widely across studies. A common observation was the imprecision or limited information about the method used to determine training load. This could lead to "under-dosing" or failure to provide an adequate stimulus to induce changes in the muscle's ability to generate joint torque, thereby affecting functional performance. This may be an especially important consideration when designing functional training programs, in which many muscles are involved simultaneously and the load on any particular muscle group is difficult to determine. All of the exercise programs reviewed were 10 weeks or less in duration. For people with lifelong motor disabilities, strengthening most likely needs to be included in a regular exercise regimen in order to maintain optimal musculoskeletal function throughout the life

span. Research is needed to examine physical fitness programs with durations of at least 1 year in order to make statements about long-term health benefits and adherence.

An alternate means to produce strength changes is to use electrical stimulation to artificially activate the targeted muscle or muscle groups. In view of muscle activation deficits,^{4,8} electrical stimulation is a theoretically attractive method because muscle contractions are elicited independent of voluntary control. A review of the literature, however, did not show conclusive evidence of its effectiveness in children with CP.⁴⁹ Further research is currently under way.⁵⁰ An in-depth review of this approach is beyond the scope of this article.

Muscle Strength Measurements

Muscle strength is assessed at the body functions level of the ICF. Research protocols commonly utilize isokinetic testing devices or handheld dynamometry to assess muscle strength. Strength is typically assessed by measuring joint moments, commonly referred to as "joint torque" in the clinical literature. To make comparisons among children of varying ages and anthropometric characteristics, joint torque is divided by body weight.

Isokinetic testing devices are often used in the laboratory environment and are available in many physical therapy clinics. These devices offer the advantages of precise stabilization of the patient and the limb segments being assessed as well as quantification of the moment arm distance from the location of force exertion. Strength can be assessed for different speeds of movement (isotonic and isokinetic) and different types of contractions (isometric, concentric, and eccentric) but most frequently is reported for isometric and concentric conditions. Van den

Berg-Emons et al⁵¹ found high test-retest reliability using the Spearman rank-order correlation (r_s) for isokinetic testing of the knee extensors of children with CP at slower (30°/s, $r_s=.71$) but not higher (60°/s, $r_s=.55$, and 120°/s, $r_s=.42$) speeds of movement. Good reliability ($r_s=.90-.95$) was reported at all speeds for a control group of subjects without CP. In contrast, Ayalon et al⁵² reported a high level of reliability (intraclass correlation coefficients [ICC]=.95-.98) for knee extensors and flexors of a group of children with CP at 90°/s for a total of 8 measurements (4 repetitions per session, taken 1 week apart). Possible explanations for the differences in reliability included the fact that van den Berg-Emons et al examined the difference between 2 tests performed on the same day with an extensive testing protocol that may have introduced fatigue for the subjects with CP. Age may have been an additional factor because the subjects in the study by van den Berg-Emons et al were younger (mean=8.7 years, range=7-12) than those in the study by Ayalon et al (mean=11.1 years, range=9-15).

Limitations of isokinetic testing devices include the time required to test multiple joints, cost of the equipment, and lack of portability. In contrast, handheld dynamometers are less expensive, lightweight, portable, and easy to use. The force transducer is placed on a distal aspect of a limb segment while the patient exerts a maximum isometric contraction at the proximal joint. Disadvantages include a reduced ability to stabilize the patient, the potential of the opposing strength of the examiner contributing to the measured force,⁵³ strong subjects exceeding the upper limit of the device,⁷ and the need to precisely measure the distance from the transducer location to the center of the joint being tested when calculating joint torque.

Agre et al⁵³ tested a dynamometer under laboratory conditions and found that, when the force was not applied in a precise perpendicular direction, readings were inaccurate and offered a plausible explanation for the low inter-examiner reliability (Pearson correlation coefficient [r]=.49-.81) found for 6 lower-extremity muscle groups of adults without disabilities. Despite these concerns, good to high intrasession and intersession reliability of data for the hip abductors and the knee flexors and extensors were found during testing of a group of children with spastic diplegia and quadriplegia. With the exception of left knee flexion, all ICCs were .84 or higher.⁵⁴ Similarly, Taylor et al⁵⁵ concluded that there was good intersession reliability (ICC=.81-.96) for a group of children with spastic diplegic CP for repeat testing of the ankle plantar flexors, knee extensors, and hip flexors and abductors. Despite the high reliability found for group data, they questioned the consistency of individual subject performance based on the large range in confidence intervals found for these data. Further research is needed to establish the detailed protocols necessary to ensure testing reliability for intervention studies. It may be prudent to collect multiple baseline data in order to identify and exclude participants who are not capable of consistent performance for a particular outcome.

All of the above strength testing protocols required that the child exert maximal effort in a consistent manner across multiple testing sessions. Research was focused on children with fairly mild CP who were able to walk and follow verbal directions and possessed the motor control and strength required for testing protocols. Very young children and those with intellectual impairments, substantial muscle weakness, or severe movement disorders may not be capable of providing this level of coop-

eration and effort. Methods to determine muscle cross-sectional areas and volume, such as magnetic resonance imaging (MRI),^{4,50,56} are promising because they reflect the muscle's force production capability but eliminate the need for the participant's cooperation. Using MRI, researchers⁵⁶ reported hemiplegic limb thigh and calf musculature volumes that were 84% and 72%, respectively, those of the nonhemiplegic side of children with CP. Although changes in muscle volume due to an intervention are reflective of increased strength using this method, the ability of the participant to use this strength in a functional manner is not captured. Assessments performed at the activities level of the ICF, such as the GMFM, are more reflective of a child's functional strength. However, other features of the movement disorder, such as balance, contribute to the child's overall performance; therefore, not all gains in strength may be apparent at this ICF level. Measuring the effect of strength gains on participation is important but even more problematic because environmental and personal factors play a pivotal role.

Muscle Strength Summary

Muscle strength can be improved with exercise in children with CP when the load is sufficient. An increase in strength appears to have a positive influence on activity. The extent of this influence depends on multiple factors, including the duration of the program, the degree of weakness compared with the strength required for the target task, and the coexisting impairments. Impairments such as joint contractures and spasticity, which potentially limit the effectiveness of training, should be addressed to maximize functional gains. Current treatment approaches include physical therapy interventions aimed at minimizing joint contractures, serial casting, medications (botulinum toxin, ba-

clofen), and surgery. The combination of resistance training with surgical and medical treatments is another understudied area. To date, research has been focused on children with the spastic form of CP. Little is known about the effects of resistance training for children with other types of movement disorders such as athetosis, dystonia, ataxia, and hypotonia.

Cardiorespiratory Fitness Importance of Cardiorespiratory Fitness for Children With CP

There is strong scientific evidence that youth with low physical activity and fitness levels and high body fat levels are more likely to display additional risk factors for cardiovascular disease such as elevated blood pressure and serum cholesterol levels.⁵⁷⁻⁵⁹ Conversely, an expert panel recently convened by the Centers for Disease Control and Prevention concluded that daily participation in developmentally appropriate, moderate to vigorous physical activity lasting 60 minutes or longer can reduce body fat, encourage weight loss, and improve aerobic fitness in youth aged 6 to 18 years without disabilities.⁶⁰ The few studies available also suggest that some degree of positive association may exist between physical activity and various indexes of mental health, including anxiety, depressive symptoms, and physical self-concept.⁶⁰

Evidence of Reduced Cardiorespiratory Fitness

Limited research has indicated that children with CP display low levels of cardiorespiratory fitness, as evidenced by a reduced peak $\dot{V}O_2$ or a higher submaximal energy demand of walking.^{10,11,13} These findings are concerning because reduced cardiorespiratory fitness may contribute to poor general health. From a functional perspective, children with CP have difficulty performing purposeful and efficient physical movements

for many reasons, including weakness, abnormal muscle coactivation, involuntary movement, poor selective voluntary motor control, spasticity, contractures, and decreased balance.^{13,19-21,61} These impairments can limit a child's ability to play and exercise at intensities necessary to develop cardiorespiratory fitness. Fatigue, commonly reported by people with CP, is thought to be a result of using an abnormally high percentage of their peak energy resources during physical activities.^{13,62} For children with CP who are able to walk, the locomotor energy demands increase with age,⁹ making it difficult to sustain their walking endurance as they transition into adolescence and adulthood.

Reductions in cardiorespiratory fitness in children with CP have been documented by outcome measurements at the body functions and structures^{9-13,16,63-65} and activities^{13,15,16,66} levels of the ICF. At the body functions level, cardiorespiratory fitness is measured by determining the amount of energy expended during movement and exercise. The most common method of assessing cardiorespiratory fitness in clinical laboratories is indirect calorimetry, wherein heat production by the body is estimated from oxygen use. With this technique, expired air is collected using a metabolic data collection system, and gas samples are analyzed for volume and for oxygen and carbon dioxide content. Standardized equations then are used to calculate the volume of oxygen consumed.⁶⁷ Typical outcome data include gross $\dot{V}O_2$ expressed relative to: (1) body mass and time (in milliliters per kilogram per minute) or (2) body mass and distance traveled (in milliliters per kilogram per meter or milliliters per kilogram per kilometer). Oxygen uptake also can be expressed in net terms by subtracting resting $\dot{V}O_2$ values.

Using this method, research has shown that children with CP display excessive energy expenditure values for a given speed of walking.^{9,11-13,64,68} Estimates of energy use in children with CP range from 2 to 3 times higher than values for children without disabilities when walking at comfortable overground speeds^{9,64} and increase with level of walking disability.⁶⁵ Norman et al⁶⁴ documented energy expenditure in 10 children with spastic diplegic CP and 15 children without disabilities. At a lower average walking speed (41.3 versus 66.7 m/min), $\dot{V}O_2$ (19.7 versus 6.3 mL/kg/min) and oxygen cost (0.55 versus 0.09 mL/kg/m) were significantly elevated ($P < .05$) in children with CP versus controls. Other authors¹¹⁻¹³ have reported similar results for treadmill walking. Morgan et al¹¹ evaluated children with spastic hemiplegic CP and children without disabilities during treadmill walking at 40.2, 53.6, and 67.0 m/min. Although $\dot{V}O_2$ was higher at all speeds for the children with CP, significant differences were noted only at the 2 highest speeds. Rose et al¹² found that the average maximum treadmill walking speed for a group of children with CP (56 m/min) was less than half that of children without disabilities (122 m/min). Oxygen uptake per meter walked was an average of 280% higher for children with CP.

In addition to increased energy demands during walking, lower levels of peak energy reserve have been reported in children with CP (ie, significantly lower mean peak $\dot{V}O_2$ levels) compared with children without disabilities during treadmill walking.^{10,13} Walking at 50 m/min, children with CP exercised at a relative intensity of 53.5% of maximum $\dot{V}O_2$ compared with 22.5% for controls.¹³ Adults with CP may exhibit even lower levels of cardiorespiratory fitness. Fernandez et al⁶⁹ measured peak $\dot{V}O_2$ with a metabolic cart during bicycle ergometry, treadmill ex-

ercise, or arm crank exercise. Men with CP exhibited 23% to 45% lower levels of cardiorespiratory fitness compared with adults without disabilities, and women with CP exhibited even lower levels (21%-61%). Further study is needed regarding sex and age differences in fitness levels for individuals with CP.

Although the above research demonstrated reduced cardiorespiratory fitness within the laboratory, less is known about the daily physical activity levels of children with CP at home, at school, and in the community. More recently, data recording devices have been developed that provide greater insight. Pirpiris and Graham¹⁵ reported that children with CP spend more time sitting compared with their peers without disabilities. An inverse relationship was observed between severity of disability and time spent in an upright position. The average upright time was 5.6 hours for children without disabilities versus 5.1, 2.5, and 0.5 hours for children with spastic hemiplegia, spastic diplegia, and spastic quadriplegia, respectively. Mean upright times for children with CP were significantly lower ($P < .01$) than those for children without disabilities. A study by Bjornson et al¹⁴ showed that, once upright, children with CP took significantly fewer steps per day (4,244) compared with children without disabilities (6,740) ($P < .001$). The Gross Motor Functional Classification System (GMFCS)⁷⁰ is a descriptor of mobility function at the activities and participation levels of the ICF. Children who walked independently at GMFCS level I (without restrictions) averaged more steps per day compared with children who walked at levels II (with restrictions) and III (with assistive devices) ($P < .001$).¹⁴

Other researchers^{16,63,71} have reported low daily physical activity levels for children with CP based

on physiological measurements. Van den Berg-Emons et al¹⁶ examined daily physical activity using metabolic markers (a doubly labeled water technique) in 10 children with CP compared with 10 children without disabilities. The children drank a water solution containing isotopes (¹⁸O and ²H) at the beginning of a typical school day. Urine, collected at the end of the day, was analyzed to determine excretion levels of these markers. The ratio of total daily caloric expenditure to sleeping metabolic rate was significantly lower in children with CP. The advantage of this method for estimating total daily energy use is that it did not encumber the participant with the extensive equipment required for expired air analyses. However, it is expensive, and variations in physical activity throughout the day cannot be captured. In a separate publication,⁷¹ this research group reported that physical activity levels based on heart rate data, collected throughout the day, demonstrated good correlation with those based on metabolic data at the group level of analyses ($r_s=.88$). This method was used in a subsequent study,⁶³ and significant correlations, determined using linear regression analyses ($P<.05$), were found between daily heart rate levels and net $\dot{V}O_2$ at treadmill speeds that were 60% ($r=-.82$) and 75% ($r=-.70$) of free walking speed but not at 90% of free walking speed ($r=-.30$).

Cardiorespiratory Fitness Interventions

Aerobic exercise training has been shown to increase cardiorespiratory fitness in youth without disabilities, as evidenced by an improvement in maximum $\dot{V}O_2$ ^{72,73} and a reduction in the submaximal energy demands of locomotion.⁷⁴ Although there have been few well-designed intervention studies aimed at improving cardiorespiratory fitness in children with CP, preliminary evidence indicates

that gains are possible.⁷⁵ An aerobic exercise intervention was designed for 22 children and young adults (mean age=14 years) with CP (12 with spasticity, 10 with dyskinesia) whose impairments ranged from mild to severe.^{75,76} Exercise frequency consisted of 3 sessions per week, each lasting 20 minutes, for durations of 1.5 to 16 months. The primary intervention was lower-limb cycling. Children with severe disabilities who could not use an adapted bicycle were positioned prone on a board with wheels, which they propelled with their arms. Outcomes included heart rate, $\dot{V}O_2$, and blood hemoglobin. Oxygen uptake at a given heart rate increased with training by at least 10% to 25% in all except 2 participants. In addition, a positive relationship ($r=.68$) between duration of training and improvement in $\dot{V}O_2$ was found. Total blood hemoglobin, reported for 5 participants, improved in a similar fashion. Although this study^{75,76} lacked controls and a standardized protocol, it featured innovative aerobic training and data collection methods for children with severe physical impairments.

An aerobic exercise intervention designed for adults with CP also led to an improvement in $\dot{V}O_2$. This 8-week, twice-weekly cardiorespiratory fitness program was implemented for 7 adults with mild CP who were ambulatory and using a stationary ergometer (Schwinn Airdyne[†]) propelled by lower-extremity cycling and upper-extremity push-pull movements.⁷⁷ A significant increase ($P<.05$) of 12% in peak $\dot{V}O_2$ was observed when participants were tested with the Schwinn Airdyne, but no change in peak $\dot{V}O_2$ was detected when participants were tested using an arm crank ergometer. In this case, motor learning and improved neuromuscular coordination due to train-

ing may have increased efficiency. Studies are currently under way to further investigate the efficacy of a stationary cycling intervention in children with CP.⁷⁸

More generalized sport and exercise programs have demonstrated improved cardiorespiratory fitness in children with CP.^{79,80} In one study,⁸⁰ an 8% improvement in peak $\dot{V}O_2$ was shown in an exercise group of children with CP who performed individual or group games followed by mat exercises or swimming (in the summer months) when compared with a group of children who either chose not to exercise or stopped exercising early in the study. The effects of a 9-month, 4-times-weekly, school-based sport and exercise program were examined in a group of children with CP. This program included cycling, swimming, wheelchair propulsion, running, and mat exercises. Subjects were matched and randomly assigned to an exercise group ($n=10$) or a control group ($n=10$).⁷⁹ A significant improvement ($P<.05$) of 35% in peak $\dot{V}O_2$ was observed for the exercise group when compared with the control group, and an increase in fat mass was noted in the control group over this same time period.

Darrach et al²⁴ included endurance exercise as one component of a community physical fitness program for 23 participants with CP between 11 and 20 years of age. Aerobic dance routines were designed to ensure a cardiovascular workout routine with minimal balance requirements. The majority of subjects were able to attain heart rates above 145 bpm during exercise. Although heart rate-based measures of cardiorespiratory fitness during stationary cycling did not change as a result of this intervention, a significant improvement ($P=.006$) in the physical appearance subscale of the Self-perception for Adolescents Profile was reported.

[†] Pacific Cycle Inc, 4902 Hammersley Rd, Madison, WI 53711.

Other community physical fitness programs have demonstrated positive changes in indirect estimates of submaximal aerobic fitness for children with disabilities, including those with CP.^{35,36} Community physical fitness programs with physical therapy guidance may be an ideal method to transition children from individual physical therapy sessions to lifelong physical fitness programs.

Cardiorespiratory Fitness Measurements

Laboratory evaluations allow for greater standardization and accommodation within the testing environment, but differences in experimental protocols and outcome measures can hinder meaningful comparisons across studies. Therefore, it is important to consider both within- and between-day stability of cardiorespiratory fitness variables. Because many exercise protocols require subjects to perform novel tasks, such as treadmill walking, accommodation to the laboratory setting is important to minimize excessive energy expenditure due to anxiety and lack of familiarity with testing equipment and procedures.⁸¹ Energy expenditure should be measured during "steady state," which occurs after at least 2 minutes of exercise performed at submaximal intensity.¹³

Although measurement of $\dot{V}O_2$ is considered the gold standard for the evaluation of energy expenditure, these measurements are not easy to obtain in a clinic environment. Therefore, researchers have studied the relationship between direct measurement of $\dot{V}O_2$ and indirect outcomes that are easier to obtain.^{12,64,81}

The energy expenditure index (EEI),¹² initially described as the physiologic cost index,⁸² is a measure of heart rate normalized to walking speed. Both indexes are calculated by subtracting resting heart

rate from walking heart rate and dividing the result by walking speed. Rose et al¹² found a high correlation ($r=.84$) between heart rate and $\dot{V}O_2$ during treadmill walking. Although data from the study by Norman et al⁶⁴ support these findings for overground walking, there is some disagreement in the literature as to the validity of data for this measure.^{13,81} For instance, Keefer et al⁸¹ compared measurements of $\dot{V}O_2$ with EEI in 13 children with hemiplegic CP during treadmill walking. After 2 practice sessions, $\dot{V}O_2$ and EEI data were collected during walking at 3 different speeds (0.67, 0.89, and 1.12 m/s). When individual data were analyzed, a moderate association ($r=.64$) was found between net $\dot{V}O_2$ and EEI at the highest speed, but not at the lower speeds. An unmatched response pattern between net $\dot{V}O_2$ and EEI also was observed for many participants. Based on these findings, the authors recommended caution when substituting heart rate for more direct measures of energy expenditure.⁸¹

The accuracy of this method has been questioned by other investigators,^{83,84} who assessed overground walking in children with CP. These concerns reflect the fact that heart rate may be affected by factors other than $\dot{V}O_2$, such as anxiety or anticipation of exercise, particularly at rest and low levels of activity, as well as climatic stress, dehydration, fever, various diseases, and medications.⁶² As intensity of exercise increases, however, the relative importance of anxiety generally lessens.

Given these potential limitations, Rose et al⁶² suggested that an adjusted EEI value that is based on only exercise heart rate may be a better indicator of walking energy use in children with CP. Ijzerman and Nene⁸³ reported a decrease in within-subject variability when baseline heart rate was not subtracted

from exercise heart rate. Few studies have used EEI as an outcome measure following intervention. Butler et al⁸² reported a positive change in a small group of patients following orthotic intervention. One study³⁶ demonstrated an improvement in EEI following a community physical fitness program that included children with CP. Conversely, 2 other studies^{24,32} did not show a significant change in children or young adults with CP at the conclusion of physical fitness programs.

Functional tests of walking and running endurance are indirect measures of cardiorespiratory fitness at the activities level of the ICF. Results from these tests may be more representative of a child's ability to participate in play and sport activities than clinical measures of cardiorespiratory fitness derived from treadmill evaluations. Outcomes include the distance the child is able to cover in a given time frame, the time needed to traverse a given distance, or average speed.

The 600-Yard Walk-Run Test^{85,86} is a standardized physical fitness test developed for school-aged children. In this test, children are asked to complete a 600-yd (548.6-m) distance as quickly as possible by running or walking. Fernhall et al⁸⁶ used this test for children with intellectual disabilities, who could not be expected to tolerate physical education test batteries that include distances of up to 1 mile (1.6 km). They found a high correlation ($r=.80$) with laboratory measures of peak $\dot{V}O_2$. Chen et al⁸⁵ examined the ability of 46 children with spastic diplegic CP (GMFCS levels I-III) to perform this test. All except 5 children (4 at GMFCS level III, 1 at GMFCS level II) were able to complete this distance within a 15-minute time limit. Children exhibiting greater functional mobility (lower GMFCS levels) completed the test in less time.

Andersson et al⁸⁷ examined the reliability of data for a 6-minute walk test in assessing walking endurance in 25 adults with CP. The test was performed on 4 different days over a 2-week period. A learning effect was detected, such that a significant increase ($P < .01$) in speed was observed between test 1 and tests 2, 3, and 4, but not among tests 2, 3, and 4 ($P > .33$). The authors determined that an improvement of 40 m is required to be certain that an actual change has occurred following intervention and recommended the performance of a practice test prior to collecting baseline data. There is very little research examining walking or running endurance in children with disabilities following a physical fitness intervention. Thorpe et al³² used a 3-minute walk test to assess walking speed in 7 children following an aquatic exercise program. Although a significant difference was not found in this small sample, an improvement was observed in 6 children, with a mean increase of 9 m/min.

Reports of perceived exertion have been used to assess a child's effort level during testing⁸⁸⁻⁹¹ and exercise.⁹² Reliability between self-rating and physiological responses to walking or running⁸⁸⁻⁹⁰ and cycling⁹² has been reported. In these tests, the child is asked to select written verbal cues or pictures that describe varying levels of effort.^{88-90,92,93} For example, The Children's Effort Rating Table uses a scale of 1 to 10, with responses ranging from "very, very easy" to "so hard I'm going to stop."⁹³ These tools provide feedback that the exercise intensity is appropriate and within acceptable limits of tolerance.⁹² For children who have difficulty communicating by using pictures or words during exercise, external observer reports can be used.⁸⁹ Good reliability ($r = .87$) was found between external observer reports and self-report for

the Children's OMNI Walk/Run Scale of Perceived Exertion.⁸⁹ There is a lack of research examining the validity or reliability of data for these scales for children with disabilities.

Cardiorespiratory Fitness Summary

In this physically challenged population, neuromuscular impairments secondary to disease pathophysiology can produce movement patterns that are fatiguing and inefficient when compared with movement in children without disabilities. Furthermore, the muscle weakness prevalent in this population contributes to the limited reserves for sustained physical activity. These factors can lead to greater levels of physical inactivity and deconditioning and further limit physical function and mobility.⁶⁸ Given the importance of mobility to overall health, well-being, and independence in the child with CP, it is vital to: (1) identify underlying causes of excessive energy use, (2) develop effective treatments to reduce energy-wasteful movements, (3) develop physical fitness interventions for children who are unable to walk, (4) implement community exercise and sport programs to improve cardiorespiratory fitness, and (5) promote and monitor participation by individuals who have functional impairments across the spectrum of GMFCS levels. Evaluation of participation over time may be one means of identifying children at risk for losing functional mobility.

Growth, Nutrition, and Secondary Conditions

Overall growth and nutrition are important factors that must be considered in developing an appropriate physical fitness program. Abnormalities of growth and development are prevalent in children with CP.^{94,95} Children with CP may exhibit obesity⁹⁶ or patterns of poor growth.⁹⁵ The type, severity, and distribution of the specific movement disorder

appear to be important factors. One study found that 14% of children with CP were overweight.⁹⁶ As in children without CP, poor dietary intake was a contributing factor. Inactivity may be a major factor leading to obesity in some children with CP because their ability to participate in vigorous exercise and sport activities is considerably reduced. The effects of physical activity and nutrition counseling on weight loss for children with CP who are overweight have not been reported.

Poor growth appears to be more common⁹⁶ and a greater cause for concern⁹⁵ in children with CP. The additional caloric expenditure for exercise programs must be carefully considered and monitored in children exhibiting poor growth patterns. These children often are smaller, lighter, and thinner, with notably less body fat, muscle mass, and bone mass compared with children without CP.⁹⁵ In a study of children with hemiplegia and diplegia, approximately 30% of the participants were assessed as being undernourished and 23% had stunted growth.⁹⁶ Poor growth in children with CP has been associated with negative health factors such as low body fat,⁹⁷ increased caloric requirements,⁹⁸ feeding problems,⁹⁹⁻¹⁰¹ and excessive energy consumption.¹⁰² Inadequate nourishment in children with CP is related to increased hospitalizations, decreased participation in typical activities, and missed school.^{95,103} Hormonal changes are another factor that greatly influences growth and development; however, very little is known about the overall effect of puberty on growth and physical functioning in this population. It is known that puberty typically begins earlier and lasts longer in children with CP.¹⁰⁴ More research is needed to document the effects of these growth differences on health, physical function, social participation, and quality of life for all chil-

dren with CP and at all levels of severity.

Skeletal fragility, musculoskeletal deformity, and pain are among the most common secondary conditions associated with CP. Children with CP exhibit reduced bone mineral density relative to children without CP, and the difference increases with age.¹⁰⁵⁻¹⁰⁸ Risk factors for pathological fracture include severity of CP, previous fractures, insufficient growth, poor nutrition, use of anti-convulsants, low vitamin D levels, and alterations in pubertal and skeletal maturation.^{105,106} A recent study¹⁰⁹ showed a strong association between excess body fat and fracture rate in children with moderate or severe CP. There is some evidence that interventions, including physical therapy, passive standing devices, low-amplitude mechanical loading, calcium + vitamin D supplementation, and bisphosphonate medications, may be effective in the treatment and prevention of osteopenia.¹¹⁰⁻¹¹⁴ Interventions aimed at increasing bone density during childhood and adolescence may improve reserve and decrease risk of fracture later in life.

Musculoskeletal impairments, including joint contractures, are well recognized in CP and appear to be associated with pain in many children. Morrell et al¹¹⁵ found that 67% of children with CP surveyed reported pain. The spine, hip, knee, ankle, and foot were specified as regions with chronic pain.¹¹⁵ When designing exercise programs for individuals with CP, biomechanical alignment should be optimized. Additional caution should be taken when initiating exercise programs for children with musculoskeletal pain. Physical fitness programs that reduce joint stress, such as aquatic exercise,^{32,116} may be most appropriate in the presence of significant pain.

Barriers to Sport and Physical Fitness Participation

There are significant barriers to increasing and maintaining physical fitness through exercise and sport programs for children with CP. A paucity of accessible sport and physical fitness programs for children with physical disabilities is a major contributor to this problem. The Individuals With Disabilities Education Act¹¹⁷ requires public schools to make available a free public education in the least restrictive environment for children with disabilities appropriate to their individual needs. However, children with CP often report being sent to the library or cafeteria during times that are scheduled for recess or physical education for their classmates (consumer testimonials). In addition to a lack of formal physical education programs, physical movement about the classroom and school may be restricted. Wheelchair use may be preferred by school personnel due to space limitations and safety concerns, even for children who capable of walking with assistive devices. Integration into community sport and recreation activities after school and during the summer often is left up to the family, and there is often a lack of organized support from administrators (eg, park district directors) and volunteer coaches and parents to provide equal opportunity for children with CP. Adapted sport programs exist but are not available in every community. A systematic study of access and community barriers to physical fitness programs is vital to promoting the physical activity and well-being of children with CP.

As children mature and enter adolescence, they generally transition from formal physical therapy interventions to community physical activity and fitness programs. Barriers

to community fitness programs include safety, equipment, accessibility, resources, transportation, finances, perceptions and attitudes of people who are not disabled, and policies and procedures.¹⁷ Architectural barriers are costly to remove following construction, and often there is difficulty in interpreting guidelines, codes, regulations, and laws. Once inside the facility, most community fitness centers do not have accessible equipment for people with disabilities and lack exercise instructors who are knowledgeable about CP. Although modifications can be made to remove many of the physical barriers, emotional and psychological barriers are frequently greater obstacles to physical activity. Barriers to exercise such as time and lack of interest exist in the general population, and it is unknown to what extent these barriers are present in people with CP.¹⁷ Negative attitudes of professional staff toward people with disabilities and concern about liability are major barriers that have been reported.¹⁷ The need for assistance from caretakers or family members is problematic in terms of space and additional fees that may be levied.

There is a need to identify participation and adherence strategies to increase physical activity. Specific barriers have not been systematically identified for children or adolescents with CP. Research should address barriers from both personal and environmental contextual frameworks. Equipment design that facilitates appropriate and safe movement should be a priority.

Recommendations for Future Research

During this Research Summit, we identified the need for further research in specific areas of physical fitness. Although many of the factors contributing to physical inactivity in children with CP have been identi-

fied, few coordinated investigations have examined how physical fitness programs can ameliorate their negative effect on these children. The wide role and scope of practice have far outpaced the development of research to assess physical fitness interventions for children with CP.

Both traditional randomized clinical trials and qualitative analysis of interventions are needed to fully address the complex issue of fitness in this heterogeneous population. Additional randomized clinical trials are needed for assessment of fitness and exercise in subjects with CP, especially at the body functions and structures level. Overall, clinical trials are limited in terms of number, levels of evidence (eg, randomized clinical trials), outcome measures, and the scope of research. Methods for site recruitment of participants and the training and quality assurance of data collection and analysis efforts should be clearly identified. Additional research of higher quality and rigor is essential in order to make definitive recommendations. Multi-site studies are important to recruit a sufficient number of participants meeting study inclusion criteria. Due to the complex nature of the problem, it also is important to consider research methodology that is designed to provide systematic analysis of individual differences at a variety of levels, including the activities and participation levels. Quasi-experimental research designs and analytical observations may be useful in evaluating personal and environmental factors that can be overlooked in traditional randomized clinical trials.¹¹⁸

The small volume of research in this area has focused on children with spasticity and GMFCS classifications of levels I through III. More research is needed to identify appropriate training strategies and outcome measures for children with other move-

ment disorders, such as athetosis, dystonia, and ataxia, and a wider spectrum of functional impairments (eg, GMFCS levels IV and V). Modified outcome measures and exercise programs are needed for children with mobility impairments. The cardiorespiratory fitness levels of children who are unable to walk are not known.

In designing studies to improve various components of physical fitness, it is essential to consider other factors, such as surgical history, current medication use, and present levels of physical activity and therapy, that could conceivably influence the dependent variables of interest. It is essential that participants in control groups for randomized controlled trials have similar characteristics, particularly in terms of severity of disability and age. Some children with CP who have intellectual disabilities are unable to cope with the demands of testing regimens¹³ and may find it difficult to exercise at a high intensity.

Overuse training is a common problem for habitual exercisers in the general population, but the effects are unknown in people with CP. Secondary conditions associated with CP could increase susceptibility to overuse injuries. The effect of exercise on pain and musculoskeletal impairments is another unknown entity. Factors such as the type of movement disorder and the extent of musculoskeletal deformities are important variables that should be included in future research. Although the focus of this article is on children with CP, much insight can be gained by examining the effects of physical fitness programs for adults with CP. Do physical fitness exercise programs for adults with CP act to minimize secondary conditions and slow, or prevent, a decline in physical function during the aging process? Lastly, research must include subjects from di-

verse ethnic, racial, and cultural backgrounds to ensure that the needs of all individuals and their families are considered.

Cross-sectional studies of children with CP are important to identify baseline values for outcome measures related to physical fitness. Identification of laboratory, clinical, and field tests that are responsive to interventions is needed. Quality assurance is essential, especially for multi-site studies. Reliable and standardized procedures must be developed. Specific protocols should be established to standardize data collection methods and ensure the accuracy of the personnel performing evaluation procedures.

Specific aspects of intervention programs, such as muscle selection, mode of strengthening, speed and type of contraction elicited, and training parameters (frequency, intensity, and duration), are factors that warrant further systematic investigation and evaluation. Most exercise intervention studies reported frequencies of 3 sessions per week for durations of less than 10 weeks. Durations should be extended to 6 months or 1 year with adequate follow-up to examine outcomes and retention. The relationship between exercise intensity and outcomes is an important area that warrants more research. Although it is speculated that differences in the exercise "dose" may explain the wide variation seen in outcomes found for strengthening programs, the intensity of exercise is rarely reported. Research in the area of cardiorespiratory fitness is extremely limited, and much work needs to be done to determine safe and effective protocols.

Research efforts should consider the goals of the individual and family within their socioeconomic, cultural, and environmental contexts and promote meaningful collaborations with families. Outcome mea-

tures should assess all levels of the ICF. In particular, research should examine whether improved physical fitness enables children with CP to increase their participation and to experience greater overall well-being during their daily lives. As children become teens, they generally become increasingly self-reliant and are less likely to be involved in formal one-on-one physical therapy intervention programs. If children are successful and frequent participants in enjoyable community-based activities to promote fitness at younger ages, it is hoped that this will set a precedent for continued participation and self-motivation to be active as teens and adults. One reason for our limited knowledge in this area has been a lack of methods to measure important aspects of children's participation. Evaluation of the recently developed Children's Assessment of Participation and Enjoyment and its companion measure, the Preferences for Activities of Children, supports their construct validity.¹¹⁹ Future use is needed determine their utility as outcome measurements for physical fitness interventions.

Summary

This article emphasizes the need to promote and maintain physical fitness in children with CP to improve health, reduce secondary conditions, and enhance quality of life, as discussed in the Research Summit. Studies that address all levels of the ICF framework are needed throughout the life span and across the spectrum of functional levels. Collaborations for multi-site research were established during the Research Summit, and several studies are currently under way. Chronic disorders require lifelong interventions. Preventive and promotional health strategies are likely to be less expensive than more passive or reactive approaches and should lead to better health and greater independence for children

and adults with CP. It is hoped that children who incorporate regular exercise into their lifestyles will have a better chance of becoming adults who are happier and healthier, with fewer secondary conditions.

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Appendix 1.

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(Continued)

Section on Pediatrics Research Summit Proceedings

Appendix 1.

Continued

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