The role of fitness in health and disease: status of adults with cerebral palsy

DEBORAH THORPE PT PHD PCS

Division of Physical Therapy, Center for Human Movement Science, School of Medicine, The University of North Carolina at Chapel Hill, Chapel Hill, NC, USA.

Correspondence to Deborah Thorpe, Division of Physical Therapy, Center for Human Movement Science, School of Medicine, The University of North Carolina at Chapel Hill, 3006 Bondurant Hall, Chapel Hill, NC 27599-7135, USA. E-mail: dthorpe@med.unc.edu

LIST OF ABBREVIATIONS
ADLs Activities of daily living
HRQL Health-related quality of life
TEE Total energy expenditure

CONFLICTS OF INTEREST
The author declares no conflicts of interest.

Physical activity has significant health benefits and is positively associated with health-related quality of life and psychosocial functioning. Persons with disability are at particular risk of inactivity. For adults with cerebral palsy (CP), impaired health and function typically impede participation in physical activity, setting into motion a downward spiral of prolonged inactivity. Adults with CP may not be engaging in sufficient physical activity to produce the improvements in fitness required to experience associated health benefits. However, the literature related to physical activity and fitness in adults with CP is sparse. As more and more persons with CP lead productive lives into their golden years, it is imperative that the scientific community provide definitive information to help guide decisions related to the type and extent of fitness-related activities most beneficial to these individuals. This information will facilitate development of physical training programs that promote maintenance of function and fitness while preventing the onset of secondary conditions. This presentation will address the state-of-the-science regarding physical activity and fitness for adults with CP and how fitness training relates to physical activity and health in this population. Gaps in the evidence, as well as possible directions for future research, will be presented.

The concepts of ‘health’ and ‘fitness’ are very broad and carry unique meanings for each individual. In the Constitution of the World Health Organization, health is ‘a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity.’ Fitness is a set of attributes that people have or achieve that relates to the ability to perform physical activity. Fitness is the body’s ability to function efficiently and effectively in work and leisure activities, to be healthy, to resist hypokinetic diseases, and to meet emergency situations. The Centers for Disease Control and Prevention cite five components of physical fitness – muscular strength, flexibility, cardiopulmonary endurance, muscular endurance and body composition.

It is apparent that the concepts of ‘health’ and ‘fitness’ are interrelated. When interviewing adults with cerebral palsy (CP), it is evident that they are not free from disease nor infirmity; however, many, if not the majority, consider themselves to be in good health. At the same time, they all unequivocally agree that they could be more physically fit as it relates to function, work, and leisure activities.

Sixty percent of people with CP in the United States are over the age of 15 and have life expectancies reaching those of the unaffected population; nonetheless, longevity is still strongly related to the degree of disease severity. Secondary impairments are compromising to all persons with disabilities, as they may precipitate decreased mobility (ambulation or wheelchair mobilization) and activity levels (participation in work, recreation, and wellness). Adults with developmental disabilities are more likely to lead sedentary lifestyles and are at greater risk of having four to five chronic health conditions than are non-disabled adults. Diminished activity may in turn contribute to decreased productivity and reduced health-related quality of life (HRQL). Although there is a vast body of scientific information regarding the status of and interventions for children with CP, only recently have researchers begun to focus on adults with CP.
Although CP is the fifth leading cause of activity limitation in the United States and one of the most costly, research is just beginning to gauge the health status of adults with CP and to explore how developing secondary impairments interact with the aging process in this population of adults. Maruishi et al. investigated the independent effects of muscle strength and muscle tone in adults with CP. They found that topography of motor deficits, strength, muscle tone, and deformity affected the ability to complete activities of daily living (ADLs), but that weakness and muscle tone independently affected ADLs. Their recommendation was to attempt to manage these two problems independently. Only one study has attempted to characterize total energy expenditure (TEE) in adults with CP. Johnson et al. used the doubly labeled water technique to measure TEE in 30 adults ranging in age from 19 to 57 years. They found that TEE was highly variable in adults with CP, that ambulatory status was a significant predictor of TEE, and that the contribution of resting metabolic rate and of energy expended in physical activity to TEE differed between ambulatory and non-ambulatory participants. The wide variation in energy expenditure during physical activity in adults with CP suggests that traditional methods used to estimate energy requirements are not applicable to this population.

Several investigators have established that adults with CP may experience expected age-related changes earlier in life than their non-disabled peers. A disabling condition such as CP frequently causes a ‘cycle of deconditioning’ in which physical function deteriorates, followed by a further decrease in physical activity level, and a cascade of increasing functional decline. For a person with CP, changes inherent in the aging process are potentially deleterious to their ability to function; however, there is little evidence to determine which specific elements of health status are most important for maintaining independent function in persons with physical disability. For example, a person with CP may have worked for many years to develop independent balance and ambulation, only to have age-related musculoskeletal changes compound the effects of secondary impairments related to CP, such as decreased flexibility and strength. These escalating effects may disrupt the equilibrium of the system, compromising balance abilities and precipitating injurious falls. To accurately assess the health status of adults with CP and other physical disabilities, aging must be redefined as a process by which function begins to decline rather than as a point reached on the lifespan continuum.

In 2005, during her presentation at the third national Symposium on Translating Evidence into Practice (III STEP), Dianne Damiano, a leading researcher and author on strength training for persons with CP and an avid proponent of replacing treatment approaches that have only marginal positive effects with evidence-based exercise protocols, emphasized the importance of ‘practice’ and ‘physical activity’ for the physical, cognitive, social, and emotional health of persons with CP and other central nervous system disorders. She noted that the evidence provided by neurorehabilitation research suggests that we are far from approaching the human limits for physical and neural recovery in many disorders. She challenged the rehabilitation community to promote more active and intense training protocols for persons with CP.

This paper addresses the state of the science regarding physical activity and fitness for persons with CP and describes how fitness training relates to physical activity and health in this population. In addition, gaps in the evidence, as well as possible directions for future research, will be presented.

**STATE OF THE SCIENCE**

**Physical activity**

Physical activity is the culmination of all the activities that one pursues. For the able-bodied population these activities are divided between those that typically require less energy (ADLs) and those that require more energy, such as fitness and leisure activities. However, for persons with disabilities, activities such as ADLs could require maximum energy expenditure. Persons with disability are at particular risk of inactivity. Children with CP are less active than their non-disabled peers and have a lower maximum oxygen consumption (VO2max), muscular endurance, and peak anaerobic power. Often weakness, a primary impairment in persons with CP, prohibits participation in vigorous cardiovascular conditioning programs. Cooper et al., in a study on physical activity and health among people with disabilities found that activity level was highly correlated with years of survival in adults with CP.

Physical activity has significant health benefits and is positively associated with HRQL and psychosocial functioning. Rimmer has reported at length on the impact of physical activity on health and function in persons with disability. He states that in a population that is aging with disabilities, changes in function may be related to the lack of, or a decrease in, physical activity and a reduction in physical fitness. It is apparent that a small change in strength or endurance can negatively affect levels of independence, and there is good evidence linking physical activity with functional status and overall health. Impaired health and function impede participation in physical activity, and a downward cyclical pattern is established with ensuing prolonged physical inactivity. Consequently, physical activity is being targeted as an intervention strategy for older adults with physical disabilities.
Most of the evidence that exists related to physical activity in persons with CP, pertains to children.\textsuperscript{19,32–38} The constructs of both ‘activity’ and ‘performance’ are proposed to measure the general component of activity. Bjornson et al.\textsuperscript{32} emphasize that historically, physical activity has been measured by clinical measures that assess only ‘capacity’ of the child. Very few activity ‘performance’ measures exist that measure activity in the child’s natural environment. Moreover, assessment of physical activity has usually been via survey (self- or parent-report). Only recently has work been done with accelerometers\textsuperscript{32,35} that will provide true quantitative data on physical activity ‘performance’ from natural environments for children with CP.

Only a few studies\textsuperscript{5,39–41} have been published on physical activity levels of adults with CP. Turk et al.\textsuperscript{5} found that the health status of adult females with CP had a negative effect on exercise participation. Participants reported that pain (84%), musculoskeletal conditions (59%), and bowel and bladder problems (56%) limited their participation in exercise. In spite of these health problems, 83\% of the 63 community-dwelling women with CP in the study\textsuperscript{5} engaged in at least one physical activity, including swimming, walking, weight lifting, and using exercise equipment.

All investigators concluded that there is evidence that adults with CP might not be engaging in sufficient physical activity to produce the improvements in fitness that would be required to experience associated health benefits.\textsuperscript{24} All studies, with the exception of that of Van der Slot et al.\textsuperscript{40} used survey methods to collect data on physical activity. Van der Slot et al.\textsuperscript{40} measured physical activity with an Activity monitor (AM; Temec Instruments BV, Kerkenarde, the Netherlands) for 48 hours in 16 participants with hemiplegic CP (Gross Motor Function Classification System [GMFCS] level I) and 16 healthy age- and sex-matched peers. The authors found no differences between groups in the level of everyday physical activity and in most life areas; these participants participated to a similar degree as their healthy comparisons. From these data, we might conclude that it is the more severely impaired adults with CP (GMFCS levels II–V) who have severe activity limitations that might facilitate the development of secondary conditions leading to decreased health benefits.

**Fitness**

In 2003, the American Physical Therapy Association’s 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 CP. In 2004, with funding from the Section on Pediatrics, the Research Committee convened a summit with the purpose of fostering research in the area of physical fitness in children with CP. The findings of this summit reinforced the fact that there is a great need for more research on fitness in persons with CP, addressing all levels of the International Classification of Functioning, Disability, and Health framework across the lifespan and all across all functional levels.\textsuperscript{42}

Fitness can be divided into five categories: muscular strength, aerobic fitness, muscular endurance, flexibility, and body composition. This paper will address mainly the components of muscle strength and cardiorespiratory fitness.

**Muscle strength**

Most of the literature regarding strength training has been done in children with CP. A review article by Dodd et al.\textsuperscript{43} included 11 studies on land-based strengthening in children with CP and concluded that strength can be increased with training without adverse affects on spasticity and flexibility. The majority of the empirical studies cited in this review article reported strength increases, with two studies reporting increases in activity level and one study reporting an increase in self-concept. Additional work in the area of muscle strengthening in children with CP\textsuperscript{44–51} reflects varying degrees of successful results related to strength-training programs for children with CP.

Work has begun to validate the use of water as an optional exercise medium to land in this population.\textsuperscript{52–55} Utilizing the beneficial properties of water, such as buoyancy, neutral warmth, and variable resistance and elimination of the negative effects of gravity, persons with CP can optimize their full movement potential in an aquatic environment while maintaining joint integrity. Thorpe et al.\textsuperscript{54} investigated aquatic-based strengthening in seven children with CP, ages 7 to 13 years. Children participated in individualized aquatic exercise sessions three times per week for 10 weeks. They were assessed pre-, post- and at 11-weeks postintervention. Significant improvements were found in the walking dimension of the Gross Motor Function Measure (GMFM)\textsuperscript{56} and the Timed Up and Go tests.\textsuperscript{57} Trends were observed toward increasing overall lower extremity muscle strength and gait velocity.

**Cardiorespiratory fitness**

Unnithan et al.\textsuperscript{58} noted a gap in the evidence for training studies in children with CP. They identified many studies looking at single-specificity training (either strength or aerobic capacity), but no studies addressing interval training, such as those cited in the literature on typically developing adults.\textsuperscript{59} They designed a 12-week interval training program for adolescents with CP, ages 14 to 18 years, who
represented levels II and III on the Gross Motor Function Classification Scale (GMFCS). Participants trained three times a week for 12 weeks for 70-minute sessions, completing a combination of aerobic and strength interval training activities and games. As the participants’ physical fitness improved, the intensity of the training sessions (as measured by heart rate) was increased to a final level of 75% of age-predicted maximal heart rate by the end of 12 weeks. The authors concluded that significant improvements in work economy resulted and stated that this may have implications for increasing physical activity levels in children with CP and warrants further research.

A few studies have addressed progressive resistive strength and aerobic training in adults with CP. Two of the studies were done on elite athletes with CP and did not establish a baseline prior to the intervention. However, Andersson et al. did have a control group and put 10 adults (age range 25–47y) through a twice-weekly, 60-minute, 10-week progressive strength-training program that emphasized the lower extremities. The load for each position was standardized according to the 1 RM (one repetition maximum, i.e. the maximum weight that could be lifted in one repetition) method and set at 70% of 1 RM. Each participant then performed 10 repetitions in each of three sets with this weight. Results indicated significant improvements in muscle strength, walking velocity, and gross motor standing in standing and walking. Taylor et al. examined whether a community-based progressive resistive strength-training program (mostly machine weights) for the limbs and trunk could improve muscle strength and functional activity in a group of 10 adults over 40 years of age with cerebral palsy who could walk independently, with or without an assistive device or self-propel a manual wheelchair at least 10m. These individuals were more support-dependent than most of the adult participants in other studies. Participants exercised twice a week for a 60- to 90-minute session. Significant improvements were seen in strength (22% for legs, 17% for arms) and timed sit-to-stand (24% reduction in time). Positive trends were seen in self-selected locomotion speed.

Thorpe and Reilly reported similar results in a case study of a 31-year-old male with CP who ambulated with loft-strand crutches. He participated in an individualized aquatic-based strength-training program three times per week for 10 weeks with an emphasis on training lower-extremity muscle groups (with exception of hip flexors), gait velocity (increase of 3m/min), and the scores for both the Standing and Walking dimensions of the GMFMS (a 30% increase).

Generally, we accept that better physical conditioning leads to better health and prevention or reduction of secondary impairments. However, it has been only recently that therapists and researchers have looked beyond the limitations of the disease and discovered that many children and adults with CP can train at near-normal intensity levels (70–85% of maximum heart rate). Thorpe recently presented data from a study in progress on 14 adolescents with CP ranging in age from 12 to 20 years within GMFCS (Palisano R, personal communication 2003) levels I–III, who participated in treadmill and aquatic exercise training. All participants trained in target heart rate zones of 75% of their maximum heart rate for 45 minutes, three times per week. Results indicated no differences between the two groups and significant improvements were noted in the walking dimension of the GMFM, gait velocity, gait distance, lean muscle mass, and strength in knee extensors and hip abductors.

The scientific community knows very little about the health status, development of secondary conditions, and effects of aging on adults with CP. A study in progress by Thorpe, which is funded by the US National Institutes of Health is investigating the development of secondary conditions and possible relationships with HRQOL and physical activity. In addition, some adults from this study are participating in a promising aquatic resistive-exercise program in a community-based setting to determine the effects on motor function, work efficiency, body composition, physical activity, participation, and HRQOL.

**SUMMARY**

There is a paucity of literature related to physical activity and fitness in adults with CP. As persons with CP continue to live normal lifespans, and strive to lead active, productive lives into their golden years, it is imperative that the scientific community provide more definitive information to help guide decisions related to type, and dosage of fitness-related activities. This information will facilitate development of efficacious physical training programs that promote maintenance of function and fitness, while preventing development of secondary conditions.

**RECOMMENDATIONS FOR FUTURE RESEARCH**

1. Identify specific types (strengthening, aerobic, flexibility), combinations, and doses of exercise and physical activity for specific disabilities and secondary conditions.

2. Carry out in-depth investigation of the benefits of aquatic exercise where movement potential is optimized and joint integrity is maintained.

3. Determine which physiologic parameters will provide the most reliable and valid information in order to discern fitness need and change.

4. Investigate the effectiveness of interval training programs (more real-to-life) that train strength and aerobic capacity at same time (on land and in the water).
(5) Identify valid methods to determine TEE in adults with CP in order to develop physical activity interventions based on individual needs that promote strengthening and aerobic conditioning but also maintain optimal nutritional status.

(6) Conduct in-depth investigations of physical activity in the lives of adults with CP and the relationships between physical activity and participation, HRQL, and psychosocial functioning.

ROADBLOCKS TO EXPANDING RESEARCH IN PHYSICAL ACTIVITY AND FITNESS

(1) Heterogeneity of the population.

(2) Lack of valid and reliable measurement tools to assess variables such as strength, flexibility, physical activity, and participation.

(3) Ability to translate results to real-world (community) settings secondary to: (a) lack of accessibility in communities; (b) lack of public education and awareness to this population and their abilities (health clubs); and (c) lack of funding for support personnel required to assist with more severely impaired adults.

OPPORTUNITIES TO EXPAND RESEARCH IN FITNESS

(1) Community health clubs are becoming more open to having adults with developmental disabilities as members (personal trainers are being certified) so research is needed to help direct physical training programs.

(2) Federal funding agencies in the United States such as the National Institutes of Health, the Centers for Disease Control and Prevention, and the National Institute on Disability and Rehabilitation Research are very interested in funding aging research. They recognize that adults with developmental disabilities face similar, if not more devastating, age-related changes, and they see the need for interventions to protect function and prevent secondary conditions.

REFERENCES


55. Ozer D, Nalbant A, Aktop A, Duman O, Keles I, Toraman NF. Swimming training program for children with cerebral palsy: body perceptions, problem behaviour, and


