Aerobic fitness and lifestyle with non-exercise physical activity in adults with cerebral palsy

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Abstract This review article explores how to keep individuals with cerebral palsy healthy all their life. Many of these individuals live very sedentary lives, remaining at home after leaving school or completing childhood rehabilitation programs. Aerobic fitness in such individuals is known to decline rapidly in adulthood. Therefore, in order to keep aerobic fitness from falling into decline, the physical activities and aerobic fitness of these individuals must be appropriately and rigorously managed. The authors recently showed the validity of submaximal exercise testing and provided some findings that would contribute to improving aerobic fitness in these individuals. These studies indicate that even NEAT (Non-Exercise Activity Thermogenesis) or brief and frequent daily physical activity, rather than just conventional exercises or sports, could improve aerobic fitness. However, other studies recommend that these individuals engage in exercises or sports that they may not be interested in or that require facilities or occasions that are inaccessible. In conclusion, in addition to a standard exercise regimen, realistic and tangible interventions for daily physical activities need to be made available to individuals with cerebral palsy.

Keywords: physical activity, fitness, cerebral palsy

Introduction

Individuals with cerebral palsy manifest various disabilities that initiate in childhood and continue throughout life1). Medical treatment alone is insufficient to alleviate these disabilities as suggested by the concept of the International Classification of Functioning, Disability and Health (ICF)2). The present version of ICF was revised by the World Health Organization (WHO) in 2001, when the model of disability included the terms “health condition,” “environmental factors,” and “personal factors” and the terms “disability” and “handicap” were replaced by “activity” and “participation,” respectively. Therefore, the current version of the ICF now provides a comprehensive and suitable model for addressing the manifold problems faced by individuals with various disorders including cerebral palsy. It is especially important that the present ICF model includes “health condition” as part of its definition. In the past, the health condition of adults with cerebral palsy was neglected3-7). Aerobic or cardiorespiratory fitness is important for participation in activities of daily life because accomplishment of these activities requires a continuous aerobic supply of energy through the cardiorespiratory system8,9). In adults with cerebral palsy, aerobic fitness can play an important role in health conditions. Aerobic fitness is one factor relating to physical fitness. Other factors include flexibility, muscle strength and power, coordination, and muscle endurance8). Flexibility, muscle strength and power, and coordination may be worked upon in a hospital setting. However, aerobic fitness cannot be achieved in a hospital setting because adequate long-term fitness programs are rarely provided8,9,10). Preventive measures have been recommended by the American College of Sports Medicine (ACSM), which propose guidelines recommending moderate or higher-intensity physical exercise for at least 30 minutes almost every day to improve aerobic fitness and prevent lifestyle-related ailments8). One study observed that most healthy children took part in physical activity at > 50% of their heart rate reserve for 30 minutes or more every day11). However, most subjects did not continue satisfying the ACSM guidelines as they grew up12,13,14). In another study, <5% of ordinary adults met the ACSM guidelines15). However, concluding that ≥95% may be unhealthy or below the recommended level of aerobic fitness is inappropriate considering that physical activity can be defined in

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many ways\(^8\). Physical activity does not necessarily mean exercise. Levine coined the acronym NEAT, which stands for Non-Exercise Activity Thermogenesis\(^17\). Although NEAT has not been clearly defined\(^17\), the term is considered to include all the various daily physical activities, not necessarily intensive or prolonged, other than exercise or sports, such as housekeeping. NEAT must be included as an important part of daily physical activity. The energy consumed by NEAT has been shown to be greater than that expended during regular exercise routines recommended by ACSM guidelines\(^17,18\).

As can be seen from all these reports, an enormous amount of knowledge has been accumulated about physical activity and aerobic fitness in healthy individuals\(^5\). However, aerobic fitness has not been investigated in nonathletic adults with cerebral palsy until recently\(^3,19-22\). This paper reviews the reported literature pertinent to this topic and discusses the physical activity and aerobic fitness levels of adults with cerebral palsy.

**History of research on aerobic fitness in individuals with cerebral palsy**

Aerobic fitness of children or athletes with cerebral palsy has been vigorously investigated by Bar-Or\(^10\), Fernandez et al.\(^23,24\), Lundberg\(^25,26\), Maltais et al.\(^27-30\), van den Berg-Emons et al.\(^31-34\), and other researchers since the 1960s. These studies were based on work physiology that was mainly established by Astrand et al.\(^9\). This work physiology was originally intended to provide practical information to athletes to improve their performance. Since the 1960s, physiological studies have mostly focused on the wellness of children\(^3,10,25-28,31-54\) or athletes\(^44-54\) with cerebral palsy mainly rated to Gross Motor Function Classification System (GMFCS) level I or II. In the late twentieth century, many new remedies proliferated one after another in an attempt to recover loss of function in children with cerebral palsy. These included several empirical studies, most of which have been forgotten. However, some work physiology concerning this particular field of rehabilitation has survived because these studies provided insights into the improvement of residual function (i.e., aerobic fitness) in children or athletes with cerebral palsy rather than the recovery of lost function\(^3,10,25-28,31-54\). However, only a few recent studies by researchers like Nieuwenhuijsen et al.\(^55,56\) and Satonaka, et al.\(^39,42\) have reported on nonathletic adults with cerebral palsy rated to GMFCS level III or IV. Somehow, the fitness levels of this population are neglected after they complete their childhood rehabilitation programs\(^57\).

According to review articles on aging in adults with cerebral palsy by Turk et al.\(^58,60\), Rimmer\(^4,5\), and other researchers\(^6,41\), adults with cerebral palsy are more likely to become bedridden earlier than their healthy counterparts, possibly because of their low aerobic fitness levels\(^3\). Some studies suggest that rapid aging is common in adults with cerebral palsy\(^5,62\). These two factors combined may impede the performance of activities of daily living in this population, and this causes further deterioration of aerobic fitness, resulting in a vicious cycle\(^5,58\). Rimmer\(^3\) argued that improvements in aerobic fitness in adults with cerebral palsy were important to prevent deterioration of their health with age.

**Aerobic fitness of individuals with cerebral palsy**

Studies have revealed lower levels of physical activity in children with cerebral palsy than in age-matched healthy children\(^32,33,37,65\). In 1970, Berg published a study on aerobic fitness and training in children and adolescents with cerebral palsy\(^38\). Respiratory gas was collected using a Douglas bag, and oxygen uptake was measured during the submaximal exercise test, which predicts the maximal oxygen uptake (\(\text{VO}_{2\text{max}}\))\(^38\). After Berg’s study, no studies used the submaximal exercise test to predict \(\text{VO}_{2\text{max}}\) in individuals with cerebral palsy until 2011\(^19-22\) because this test was not validated for use in these individuals until then\(^3\). Instead, Lundberg measured \(\text{VO}_{2\text{max}}\), directly using the maximal exercise test and a cycle ergometer\(^25\) and demonstrated low aerobic fitness levels in individuals with cerebral palsy compared to age-matched healthy controls\(^26\). Bar-Or also directly measured \(\text{VO}_{2\text{max}}\) in children and adolescents with cerebral palsy; values were 10% - 30% lower than those of their healthy counterparts\(^40\). As research revealed evidence of low aerobic fitness levels in children and adolescents with cerebral palsy, van den Berg-Emons et al. began to emphasize the importance of physical training in these individuals\(^31\). Verschuren et al. showed lower levels of aerobic fitness in 7-year-old children with cerebral palsy than in age-matched controls\(^35\). In his 6-year longitudinal study of adolescents with cerebral palsy, Lundberg recommended physical activity classes after detecting lower aerobic fitness levels in these adolescents than in age-matched healthy controls\(^25\).

Some studies have examined aerobic fitness in athletes with cerebral palsy. However, athletic competitions rarely include individuals with cerebral palsy\(^44,50,52-54\). Van der Woude et al. included a few individuals with cerebral palsy in a study involving subjects with various impairments\(^51\). Bhambhani et al.\(^41\) conducted a study including only individuals with cerebral palsy, who showed good tolerance in the maximal exercise test. These above-mentioned studies aimed at investigating the effects of physical training or sports on aerobic fitness in children or athletes, including those with cerebral palsy.

Studies on aerobic fitness in nonathletic adults with cerebral palsy have been restricted to individuals who could tolerate the maximal exercise test\(^12,23,34-36,44,64\), although the aerobic fitness levels were found to be low in these study participants.
Submaximal exercise test for nonathletic adults with cerebral palsy

\( \text{VO}_{2\text{max}} \) is achieved when \( \text{VO}_2 \) reaches a plateau while an individual exerts maximal effort during the maximal exercise test. Most previous studies employed the maximal exercise test rather than the submaximal exercise test for measuring \( \text{VO}_{2\text{max}} \) in individuals with cerebral palsy rated to GMFCS level I or II\(^{,21,34,36,46,62} \). Other than the efforts by Berg\(^{39} \) as mentioned earlier, the submaximal exercise test had not been employed in this population until recently\(^{,19} \). Although various kinds of tests were validated in healthy individuals\(^{,36-49} \), the submaximal exercise test was not validated in individuals with cerebral palsy until the work of Satonaka et al\(^{20} \), who showed \( \text{VO}_{2\text{peak}} \) derived from the maximal exercise test significantly correlated with the \( \text{VO}_{2\text{max}} \) that was estimated from the 3-stage submaximal cycle ergometer test in 16 individuals with athetotptic cerebral palsy for the first time\(^{20} \). This study involved individuals with cerebral palsy mainly rated to GMFCS level III and IV. This test is based upon a linear relationship between heart rate and \( \text{VO}_2 \) above a heart rate cutoff point\(^{,70,71} \). Considering the lack of vigorous physical activity and sports in most adults with cerebral palsy, the maximal exercise test is unsuitable for them. This test may not only be impractical for use in nonathletic ordinary adults with cerebral palsy but may also be dangerous in some cases. Therefore, the submaximal exercise test provides a reasonable alternative for the measurement of aerobic fitness in this population\(^{,8,20} \). Studies using the submaximal exercise test would provide more information on aerobic fitness in adults with cerebral palsy. Such knowledge is necessary to improve the well-being of people with cerebral palsy. Many easier submaximal exercise tests can be used instead of the multi-stage submaximal cycle ergometer test\(^{,64-69} \). For example, the 20m (meter) shuttle run test evaluates physical performance and can be used as a field test; however, its results cannot be substituted for those of \( \text{VO}_{2\text{max}} \) in laboratory settings\(^{,72} \).

Till date, there are no reports on problems specific to cerebral palsy during the measurement of \( \text{VO}_{2\text{max}} \), except that the ratings of perceived exertion (RPE) are unreliable in individuals with cerebral palsy\(^{21} \). Muscle spasticity, which is one of the symptoms of cerebral palsy, may account for unreliable RPE; future studies must examine this possibility. Therefore, relying only on RPE for monitoring of exercise intensity may be dangerous, as suggested in the study by Satonaka et al\(^{21} \). According to these authors, some individuals with cerebral palsy and less exercise experience actually do not feel the intensity of physical activity even if their \( \text{VO}_2 \) is near maximum.

No evidence of the influence of spasticity or involuntary movement characteristic of cerebral palsy on cardiorespiratory function has been gathered yet. Rose et al.\(^{73} \) compared the oxygen pulse in children with cerebral palsy with that of healthy children during exercise, and found no significant difference. Moreover, there is no mention of other exercise-related abnormal reactions in the cardiorespiratory systems of individuals with cerebral palsy in other reports\(^{,9,21,22} \).

Aerobic fitness may be improved by physical activity other than training

Aerobic fitness levels in children and adolescents with cerebral palsy rated to GMFCS level I or II have been evaluated as lower than those of their healthy counterparts\(^{,23,37} \). This has been attributed to lower physical activity levels in the former population\(^{,25,37} \). This trend of less physical activity may begin after completion of childhood rehabilitation programs or leaving school\(^{,35,37} \). Individuals with cerebral palsy often have difficulty in finding jobs, and their physical activity and aerobic fitness levels may decline in adulthood. Therefore, regular exercise has been recommended for such individuals\(^{,4,24,29,34,37,62} \). However, some researchers have suggested that existing recommendations for healthy individuals to improve aerobic fitness and physical activity levels are not always applicable to individuals with cerebral palsy\(^{,4,58,74} \). There are no specific recommendations or physical activities to improve aerobic fitness levels in individuals with cerebral palsy\(^{,3,6} \). Although training and exercise may improve aerobic fitness in individuals with cerebral palsy, their aerobic fitness levels are not always low without these regimens\(^{15} \). Many individuals with cerebral palsy do not care for exercise or sports. In addition, social resources to provide exercise or sports programs for physically challenged individuals are still insufficient.

Considering that even healthy adults who exercise regularly are a minority\(^{19} \), adults with cerebral palsy who exercise daily are also probably a minority. Nevertheless, all healthy adults who do not exercise daily, do not always have low levels of aerobic fitness, suggesting that daily physical activities other than exercise may also improve aerobic fitness levels. It may also be possible that aerobic fitness in nonathletic adults with cerebral palsy may be improved by daily physical activity other than exercise. Future studies are required to provide evidence that supports this hypothesis.

Satonaka et al. found a correlation between \( \text{VO}_{2\text{max}} \) as predicted by the submaximal exercise test and the skewness of the frequency distribution of the continuous heart rate\(^{,19,22} \). When the skewness was negative, \( \text{VO}_{2\text{max}} \) was found to be favorable\(^{,19,22} \). Most participants involved in that study had positive values, indicating mostly low aerobic fitness levels\(^{,19,22} \). On the other hand, \( \text{VO}_{2\text{max}} \) was unrelated to the average values for continuous heart rate, intensity of the daily physical activity, and duration of daily physical activity\(^{,19,22} \). This study showed that brief, but frequent physical activity could improve aerobic fitness levels in this population, and that intensity and duration of physical activity are not always important\(^{19,22} \). In these
studies, the average heart rate, which is a more common parameter than skewness, was unrelated to VO\textsubscript{2max}.

The 6-h (hour) test for determining physical activity levels

Physical activity levels can be measured using heart rate changes that depend on a state of activity within the finite range of approximately 60 beats/min (at rest) to approximately 200 beats/min (at maximal effort). The frequency distribution of continuous heart rates usually includes 2 major clusters, unless daily physical activity does not include intense physical activity such as exercise\textsuperscript{19}. One cluster comes from a sedentary state while the other comes from a physically active state. When the sedentary state is dominant, the frequency distribution of continuous heart rates is skewed toward the lower heart rate. In contrast, when the physically active state is dominant, the frequency distribution is skewed toward the higher heart rate. Therefore, the skewness of the frequency distribution of continuous heart rates may reflect the activity levels of the individuals\textsuperscript{19}.

Satonaka et al.\textsuperscript{19,20,22} evaluated physical activity levels of individuals with cerebral palsy for 6 h during the daytime, when people are usually awake and physically active (6-h test), using the skewness of the frequency distribution of continuous heart rate. Continuous recording of heart rate for long periods is required to evaluate physical activity levels\textsuperscript{75}. Continuous heart rate measurement for 24 h would be necessary to estimate the total energy expenditure during an entire day. However, in order to determine trends in physical activity, continuous heart rate measurement for 6 h during the daytime is adequate when subjects are awake and physically active. This method is most appropriate to avoid sedentary periods that may alter measurements. When heart rate measurement is performed for longer periods, the sensitivity of skewness as an indicator of physical activity may decrease.

When subjects are in the active state, their average heart rate increases. However, average heart rate is not a suitable parameter for comparing individuals; because at a submaximal work level, values are smaller for individuals with good aerobic fitness than for those with lower aerobic fitness levels\textsuperscript{9,10,76}. Therefore, while the skewness of the frequency distribution of continuous heart rate may be an indicator of physical activity, average heart rate cannot be used for comparing physical activity levels among individuals\textsuperscript{19,22}. In addition, the average heart rate was not used in the measurement of NEAT\textsuperscript{18}. Taken together, the skewness of frequency distribution of continuous heart rates accurately represent individual activity levels\textsuperscript{19,22}.

Physical activity, exercise, and NEAT

Physical activity is defined as "any body movement produced by skeletal muscles that results in energy expenditure"\textsuperscript{18}. Therefore, physical activity may include various human behaviors in daily living, ranging from housekeeping and leisure activities to various kinds of jobs. Exercise is one of the subcategories of physical activity. Usually, exercise is performed intentionally for the purpose of improvement or maintenance of various aspects of physical fitness, such as joint flexibility, muscle strength and power, and aerobic fitness\textsuperscript{10}. Levine defined physical activity other than exercise as NEAT\textsuperscript{17}). Therefore, the total energy expenditure during daily life includes energy expenditure at rest, through NEAT, and during exercise, and it is calculated by the following formula:

\[
\text{kcal}_{\text{Total Energy Expenditure}} = \text{kcal}_{\text{rest}} + \text{kcal}_{\text{NEAT}} + \text{kcal}_{\text{exercise}}
\]

Since World War II, too much emphasis has been placed on exercise. However, Levine showed that the accumulation of energy through NEAT was usually greater than that through daily regular exercise routines recommended by some guidelines. Therefore, NEAT can contribute to the prevention of life-style-related diseases\textsuperscript{77,18}. Because NEAT includes various kinds of physical activities, it is often difficult to quantify. Specific parameters for NEAT have not yet been proposed. Therefore, the relationship between NEAT and aerobic fitness remains unclear. The skewness derived from the 6-h test of Satonaka et al.\textsuperscript{19,22} may be a useful parameter to quantify NEAT.

Lifestyle recommendations for aging adults with cerebral palsy

The life of individuals with cerebral palsy is very different from that of their healthy counterparts. After they leave school or complete their childhood rehabilitation programs, seclusion is not uncommon. When this is the case, low physical activity levels in addition to motor disability can rapidly deteriorate their health and physical fitness. To address this problem, more programs promoting physical activity must be provided. Medical professionals like physical therapists must be involved in this process. As mentioned earlier, the relationship between aerobic fitness and physical activity in adults with cerebral palsy is unclear\textsuperscript{4,6,56}, however, brief and frequent physical activity or NEAT\textsuperscript{17}) may be beneficial for those individuals based on the results of our 6-h test studies mentioned above\textsuperscript{19,22}). Individuals with cerebral palsy are strongly recommended to perform brief and frequent physical activity of any intensity. Physical therapists should take this into consideration while developing interventional programs for daily physical activity in these subjects.

On the other hand, the habitual behavior of individuals with cerebral palsy in childhood can affect their health and wellness in adulthood and old age. Children with cerebral palsy must not be coddled; their independence must be fostered in order to prevent a sedentary lifestyle in adulthood. Because they often receive intensive medical rehabilitation in childhood, they may view sports and ex-
exercise as leisure activities. Such individuals also may feel discouraged from engaging in sports activities because of orthopedic problems in their lower limbs. To prevent the development of a negative view of physical activity while growing up, they must be encouraged to enjoy physical activity as much as possible from early childhood\(^1\). Most recently, Terada, et al. evaluated aerobic fitness with oxygen pulse in bedridden individuals with atetosplastic cerebral palsy rated to GMFCS level V\(^7,8,79\) And, Terada, et al. showed that a year-long intervention of wheelchair dance increased aerobic fitness in these bedridden individuals\(^79\). Many enjoyable physical activities such as wheelchair dancing\(^7,79\) are ideally suited for physically challenged individuals\(^77\).

**Interpretations of VO\(_{2max}\) in adults with cerebral palsy**

Although VO\(_{2max}\) is often normalized by body mass\(^7,2,80,82\), this practice is misleading because an evaluation of cardiovascular performance should be based on total oxygen uptake\(^9\). Furthermore, Peterson, et al. showed that adults with cerebral palsy had greater intermuscular adipose tissue and greater trunk adiposity compared to healthy counterparts\(^31\). Since total body fat does not affect VO\(_{2max}\) \(^9,7,2,80,82\), VO\(_{2max}\) should be denoted as mL/min instead of mL/kg/min in adults with cerebral palsy. If it is necessary to compare VO\(_{2max}\) among individuals, it should be normalized by lean body mass\(^9\).

**Conclusions**

Aerobic fitness levels of athletes or children with cerebral palsy mainly rated to GMFCS level I or II have been vigorously investigated by many researchers since the 1960s. However, few studies have examined aerobic fitness and physical activity levels in adults with cerebral palsy. More recently, studies on nonathletic adults with cerebral palsy rated to GMFCS level III, IV, and V have been published. Some research suggested that brief and frequent daily physical activity (e.g., NEAT) may be more important, than regular prescribed exercise for 30 min, in improving aerobic fitness levels in adults with cerebral palsy. The submaximal exercise test has been validated for use in adults with cerebral palsy, and it will allow the accumulation of knowledge about their physical activity and aerobic fitness levels in the future. It is hoped that future studies will improve the well-being of adults with cerebral palsy.

**Conflict of Interests**

The authors declare that there is no conflict of interests regarding the publication of this article.

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