



# Emerging Issues in Cerebral Palsy Associated With Aging: A Physiatrist Perspective

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The population of adults diagnosed with cerebral palsy (CP) is increasing along with the survival rate of children born with the disability. Adults with CP need health services for the continued monitoring and management of their condition. Moreover, the development of additional health problems in adulthood increases the need for ongoing access to health services. Adults with CP manifest a higher rate of chronic health conditions and eventual decline in strength and functional reserve, deterioration in physical activity, increased risk of musculoskeletal complications, and gradual changes in swallowing ability. They are also reported to exhibit difficulty engaging socially and have a low health-related quality of life (QOL). However, there are a large number of adults with CP who cannot access medical services adequately and are therefore not effectively treated. To overcome these apparent challenges, we need to fully comprehend the healthcare needs of adults with CP to develop adult-focused health services. Further research is needed regarding the impact of physical activity, nutrition, sarcopenia, myeloradiculopathy, and swallowing function on QOL.

**Keywords** Cerebral palsy, Adult, Quality of life, Deterioration

## INTRODUCTION

### Population of adults with cerebral palsy

Cerebral palsy (CP) is the most common motor disability in children with an estimated prevalence of 2.6 to 2.9 per 1,000 births in the United States between 2011 to 2013 [1]. According to a study conducted in South Korea, the prevalence of CP was 3.2 per 1,000 children in 2008 [2]. Improvements in the level of medical care have resulted

in a steady decline in the incidence and severity of CP in prematurely born children since 1990 [3]. However, the population of adults with CP is increasing along with the increased survival rate of infants born with the disability [4]. A survey of individuals with CP born after 1990 revealed that 60% of the children at Gross Motor Functional Classification System (GMFCS) level V and 96% of the children at GMFCS level I or II survived until 19 years of age [5]. According to data based on the surveillance rates

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in Sweden up to 50 years of age [6], the risk of death in all age groups was consistently high in adults with CP. In a long-term study of patients with CP between 1959 and 2002, it was reported that 180 of 1,880 (9.6%) individuals died during the follow-up period. Individuals with mild-to-moderate motor impairment had a cumulative survival rate of more than 95% at age 50, compared with 74.5% among those with severe motor impairment. It was estimated that approximately 400,000 adults with cerebral palsy lived in the United States in 1995, which is gradually increasing due to medical advances and prolonged life expectancy, with an estimated number of more than 1,000,000 adults with CP in the US currently [7].

## TRANSITION TO ADULTHOOD

CP is superimposed on the dynamic process of development and aging despite being defined as static motor impairment [8]. Adults with CP need health services for the continued monitoring and management of their condition. Furthermore, the development of additional health problems in adulthood increases the need for ongoing access to health services.

Health challenges diagnosed in adults with CP differ from those in childhood and adolescence in patients with CP. Individuals with CP manifest an increased risk of death due to cancer including breast cancer and cancers of digestive and genitourinary organs [9]. In addition to ongoing needs traced to childhood, regular reviews of cardiovascular health, cervical cytology and mammography in women, access to dental care and screening is important in adults diagnosed with CP. Adults with CP have been additionally diagnosed with chronic pain and fatigue, osteoarthritis and osteoporosis, and an overall reduction in mobility [10]. According to Young et al. [11], adults with complex physically disabling conditions acquired in childhood including CP exhibit ongoing health issues and require frequent medical care. Their hospitalization rate was 9-fold higher than that of adults without disability. The pediatric and adult healthcare systems are structurally distinct in most countries, and the transition to adult healthcare occurs at 18 or 19 years old [12]. This transition is determined by age alone and not based on the patient's ability to direct their own care. Therefore, at a point when most adults are not ready to inherit further responsibility for their own medical care, they are

switched to a more independent adult-oriented healthcare system.

It is also reported that young people diagnosed with CP do not receive sufficient information prior to transitioning underscoring the need to foster knowledge and improve their skills during the transition period [13]. To overcome these apparent challenges, we need to fully understand the healthcare needs of adults with CP in order to guide the development of adult-focused health services [14]. The transition from childhood to adulthood in CP has been debated for the past 20 years with aging now emerging as a new and independent challenge. Also, the current knowledge of physiatrists among elderly adults with CP is an important issue.

## GENERAL HEALTH ISSUES

### Cardiometabolic and pulmonary morbidity

In a population-representative sample in United States, adults with CP were found to carry a higher rate of chronic conditions [15] such as diabetes, asthma, hypertension, other heart conditions, stroke, and emphysema than adults without CP. In middle-aged adults with CP between the ages of 40 and 60 years [16] and young adults with CP [17], the proportion of individuals with multiple morbidity ( $\geq 2$  among 12 chronic conditions) was high. Furthermore, obesity and a high GMFCS level were linked to increased risk of multimorbidity.

Risk factors for cardiovascular disease exist among young adults diagnosed with CP [18-20]. Age has been identified as the strongest independent predictor of vascular health [19,20], with a higher prevalence of non-ambulatory than ambulatory CP [21]. Also, fatigue [22,23] and bowel symptoms [24] were prevalent in adults with CP. According to Ryan et al. [25] in Ireland, the prevalence of metabolic syndrome was high in 55 young adults with CP with an average age of 37.5, with a prevalence of 20.5% among ambulatory adults and 28.6% in non-ambulatory adults. In a study of young adults with ambulatory CP, moderate physical activity was reported to be associated with a lower cardiometabolic risk suggesting that fitness and physical activity were important measures to reduce non-communicable disease [25-28].

### Unmet healthcare needs

Rehabilitation facilities were insufficient to manage ag-

ing in adults diagnosed with CP in spite of reduced functional abilities [29]. Surveys conducted in Sweden during the year 2001 suggested that 60% of the cohorts investigated were engaged in some kind of physical training [30]. As reported by Balandin and Morgan [31] in 1997, 20% of adults with CP have difficulties accessing medical services and 41% lack access to facilities. Furthermore, the survey also highlighted difficulties in communication and the need for external assistance. According to a recent report in South Korea [32], the medical check-up rate including private health screening, workplace health checks, medical check-ups provided by national health insurance services, and free medical check-ups in adults with CP were lower than in the total population with disabilities, with financial burden cited as the biggest factor preventing hospital attendance. According to the study, 53.2% of the individuals underwent a medical check-up in the past 2 years, while 44.2% did not [32]. Only one-third (37.0%) of individuals with CP received rehabilitation therapy in South Korea [32]. Individuals with CP reported the need for the following medical treatments: pain treatment (42.9%), additional physical therapy (35.7%), examination by a physiatrist (27.3%), orthosis prescriptions (14.3%), occupational therapy (11%), and surgery (3.9%) [32]. In addition, due to the financial burden and lack of knowledge of patients diagnosed with CP, the demand for rehabilitation services was not met in one-third of all adults with CP.

## DETERIORATION IN PHYSICAL ACTIVITY

In adults with CP, physical activity was reported to decrease with increasing GMFCS levels [33]. There is a lack of evidence supporting the efficacy of the intervention to sustain and increase habitual physical activities in children and youth with CP [34-36]. The effect of deterioration in physical activity or exercise intervention on long-term health in patients with CP has yet to be elucidated. In line with the previous review, the Cochrane study group revealed that there is low-to-very low-quality evidence supporting the benefit of physical activity in children in terms of improved gross motor function and gait speed [35]. Although few studies have examined the effects of physical activity in adulthood, increased physical fitness in young adults with spastic CP was effective in improving fatigue, mental health, and social participa-

tion without a significant change in gross motor function [23]. Although physical activity is expected to have a significant long-term impact on adults compared with pediatric populations, studies have yet to investigate the effects of physical activity in adults. A comprehensive review and further analysis of the effects of physical activity on adults with CP are needed.

## MUSCULOSKELETAL ISSUES

### Osteoporosis and arthritis

Adults with CP may be at an increased risk of hypovitaminosis D-induced osteopenia and impaired bone mass [37]. Adults with CP are more frequently deficient in vitamin D due to the use of anticonvulsants or fewer outdoor activities. Recent studies have shown an insufficient or deficient vitamin D levels in more than 50% of the adults with CP based on abdominal obesity, which is an independent predictor of lowered vitamin D levels [38]. Limited weight-bearing, inappropriate nutrition, medications (especially for the treatment of epilepsy), and other factors represent potential risk factors for the early onset of osteoporosis [10].

According to a study regarding the age-related trajectory in adults with CP, the odds ratios of osteoporosis, osteoarthritis, and rheumatoid arthritis in adults aged 30 years and older were higher than in those aged 18-30 years [39]. Multiple musculoskeletal morbidities among adults aged 31 to 40 years were 1.9-fold higher compared with adults aged 18-30 years, 4.3-fold in adults aged 41 to 50 years, and 6.1-fold higher than those above 50 years. Low bone mineral density was also observed in ambulatory adults with CP [40,41], along with frequent fragility fractures [42]. Because adults with CP are more vulnerable to osteoporosis, more frequent check-ups and active interventions are required starting at a younger age. Evaluation of body composition using dual-energy X-ray absorptiometry (DXA) or lean tissue mass is clinically important for optimal outcomes following feeding and exercise interventions [42]. In addition, interventions to increase weight-bearing or muscle mass are preferred [42]. It is also particularly important to reduce the risk of falls in ambulatory individuals with CP [42]. Since a greater risk has been reported in adults with CP, the development of a protocol for the diagnosis and treatment of osteoporosis in individuals with CP is needed.

### Sarcopenia

Botulinum toxin injections, orthopedic surgery, and neurosurgical interventions are occasionally performed in children and adolescents with CP due to muscle imbalance during their developmental period. Increasing muscle mass can be a challenge due to the lack of muscle mass when children become adults.

Adults with CP carry smaller and less dense psoas major [41], suggesting greater muscle fat infiltration, poor muscle quality, and less contractile tissue in the muscles [37]. Even ambulatory young adults with CP presented with a calf area 45% smaller than in a typically developing population [43].

Because individuals with CP show a low basal metabolic rate, obesity may occur even with a calorie-controlled diet [41]. Since premature aging associated with sarcopenia in CP may cause acute functional deficits and disabilities [44] and sarcopenia is frequently detected in adults with CP, dietary modification, nutritional supplements and exercise therapy are needed. The development of a detailed exercise protocol for adults with CP is required because increased muscle mass improves strength, functionality, endurance, and general metabolic health in individuals with CP [37]. Protein intervention stimulates skeletal muscle protein synthesis and inhibits protein breakdown resulting in positive protein balance and a net gain in muscle mass [37]. Frequent exercise with protein ingestion accelerates muscle synthesis, accretion of muscle protein, and facilitates muscle hypertrophy [37]. Regular exercise is important to improve the muscle changes associated with aging in individuals with CP [37]. Supplementation with at least 800 IU of vitamin D is effective in improving muscle strength and preventing falls and fractures, and therefore, vitamin D supplementation is also important in this population [37]. A well-designed research protocol highlighting the intervention in sarcopenia treatment is also needed.

### Pain

According to Murphy et al. [7], adults with CP aged below 50 years frequently reported cervical pain, back pain, muscle pain, joint pain, hand paresthesia, and overuse syndrome. A study demonstrating a 10-year long-term deterioration of perceived health and functioning found that pain and fatigue were the most common health challenges faced by adults with CP [45]. Another study

revealed chronic pain (lasting more than 3 months) in 75% of the adults diagnosed with CP [22]. Multiple studies revealed that the back, hip, and the lower limbs were the most common pain locations in adults with CP [10,11,22,32,45-50]. Adults with CP also manifested pain associated with contractures, orthopedic deformities, fractures, pressure from sitting on bony prominences, as well as spasticity [44]. Despite widespread recognition of pain in adults with CP, the impact of pain-related quality of life (QOL) has not been studied adequately.

However, a large number of adults with CP cannot access medical services in South Korea [32] and as a result, are not adequately treated for pain. Adults with CP need access to medical facilities for appropriate evaluation and treatment because chronic pain may result in decreased gait function [46] and reduced QOL.

### Neurological challenges associated with myelopathy

Cervical myelopathy is often induced by early degeneration of the cervical spine due to abnormal movements in adults with CP especially in adults with dyskinetic CP [51-54]. Surgical intervention has also been used to reduce exacerbation of weakness and paresthesia [55,56]. The surgical outcome was not as favorable as expected most likely due to continued and persistent neck movements after surgery.

Therefore, in order to improve the surgical outcome and to prevent the recurrence of myelopathy, dyskinetic movements should be reduced following perioperative botulinum toxin injections [52], used to reduce cervical dystonia-related pain and disability in adults with dyskinetic CP [57].

### NUTRITIONAL CHALLENGES AND DYSPHAGIA

Malnutrition has been reported in adults with severe functional disability [58]. Due to the geographic dependence and altered economic status, obesity accounts for a large proportion of the nutritional challenges reported recently [17,38]. However, further studies are needed because of the paucity of evidence supporting the effect of interventions targeting obesity and malnutrition in adults with CP.

Children with CP are reported to manifest a higher prevalence of dysphagia. Adults with CP, despite maintaining the same dietary patterns as in childhood, may

experience gradual deterioration in their swallowing and mealtime capabilities.

A qualitative study investigating swallowing difficulty in adults with CP [59] reported gradual changes in swallowing capabilities starting as early as 30 years of age. Even in adults with dyskinetic CP exposed to unrestricted diets and not previously evaluated for swallowing function, a videofluoroscopic swallowing study revealed frequent aspiration without cough reflex [60] probably due to abnormal sensorimotor integration or chronic aspiration-induced desensitization of the laryngeal airway. Considering the impact of aspiration risk on general health, the swallowing function is a challenging issue in all CP populations.

However, quantitative studies investigating the prevalence and the effect of therapeutic intervention on swallowing function in this population have yet to be conducted despite the high frequency of swallowing problems directly affecting the QOL.

## FUNCTIONAL LIMITATION

According to a study conducted in the Netherlands, 70% of young adults with CP between the ages of 18 and 22 reported challenges with activities of daily living [61] including difficulties in self-care, productivity, and leisure activities, especially involving recreation, leisure, meal preparation, and housework.

Adolescents with CP gradually exhibit a progressive decline in strength and functional reserve through adult life [37]. Prior to the age of 35 years, the ability to walk decreases in adults with CP despite acquired ambulation during adolescence. Deterioration in GMFCS levels is most evident in the late 20s and early 30s, and dependence and perceived difficulties in activity influence adults with CP [45,46,62]. It has been reported that the ambulatory function deteriorates in adulthood [7,30,63], which is likely due to new medical age-related challenges in patients with CP based on the fact that the GMFCS level remained almost stable in individuals with CP until the age of 21 years [64]. If GMFCS I-III declined to IV-V with age, it is likely that adults with CP represent a burden for their family members and caregivers, and increasingly need assistive device use. Therefore, functional limitation should be addressed in health policy regulations for adults with CP.

Limitations in functional activity were found to be a major restricting factor for social participation in young adults with CP. Although intellectual disability rather than GMFCS level in children is known to have a significant impact on social participation [65,66], there is a lack of evidence to support this finding in adults with CP. Further, work participation is restricted in adults with CP who do not suffer from an intellectual disability [67], and further research is required to encourage increased participation in society and in the workplace.

The International Classification of Functioning, Disability and Health (ICF) guidelines were developed to accurately assess the function of individuals with disabilities. The ICF Children and Youth version (ICF-CY) core set was used to measure functional limitation in children and adolescents with CP below 18 years of age [68]. The ICF core set in adults with CP has yet to be developed and the need to use ICF to accurately assess adults with CP has been highlighted [10]. Recently, there has been a move to develop the ICF core sets for adults with CP.

## HEALTH-RELATED QUALITY OF LIFE AND SOCIAL PARTICIPATION

Adults with spastic bilateral CP are reported to have difficulty engaging socially and have a low health-related quality of life (HRQOL) [69]. According to a 8-year follow-up study conducted in Canada on adults with CP, HRQOL deterioration was most evident in their late 20s and 30s [70]. However, a longitudinal multicenter study conducted in the Netherlands reported lower HRQOL in adults with CP than in populations without disability and found that HRQOL and social participation were fairly stable for many years [71].

To date, a 36-item Short Form Health Survey (SF-36) has been used to evaluate HRQOL in adults with CP [26,45,63,69,71] although they have not been validated in this population. The survey consists of physical functioning, physical role, bodily pain, general health perception, vitality, social functioning, emotional role and mental health. Each area is assigned a maximum score of 100 points. Among these items, physical functioning and physical role are reported to be low in adults with CP in the Netherlands [69]. Although the SF-36 is applicable only to people without cognitive impairment and cannot be used to evaluate individuals with intellectual dis-



abilities, it can be suggested as a useful assessment tool for adults with CP. Unmet needs for medical and rehabilitation treatment in South Korea represent a possible barrier to improved QOL and social participation among individuals with CP, and therefore, appropriate financial and technical resources are required to address the unmet needs of these populations in the healthcare system [32].

## CONCLUSION

In addition to early detection and habilitation or rehabilitation of children with CP, transition to adulthood has been highlighted as an important issue in the past 10 to 20 years. Aging in this population is an emerging issue. Physiatrists require adequate knowledge to prepare for the aging population of adults with CP and further studies are needed to investigate the impact of physical activity, nutrition, sarcopenia, myeloradiculopathy, and swallowing function in these individuals.

## CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

## AUTHOR CONTRIBUTION

Conceptualization: Bang MS. Writing - original draft: Yi YG, Bang MS. Writing - review and editing: Yi YG, Jung SH, Bang MS. Approval of final manuscript: all authors.

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